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Weight management experiences of overweight and obese Canadian adults: findings from a national survey

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Abstract

Introduction: We know little about how the 2006 Canadian Clinical Practice Guidelines for the management and prevention of obesity relate to Canadians' weight management experiences or whether these experiences reflect the recommendations in the Guidelines.

Methods: We used data from a general population omnibus survey to understand these two issues, particularly in relation to chronic disease. The survey included 23 questions related to weight management practices as well as those related to demographic characteristics.

Results: Of 2004 respondents, 33% were classified as overweight and 20% as obese. In the 12 months before the survey, 48% of overweight and obese respondents reported asking their physician about weight loss, while 30% reported that their physician advised them to lose weight without them specifically asking. With regard to the recommendations within the Guidelines, 14% of overweight and 18% of obese respondents reported having their waist circumference measured, 82% of overweight and 87% of obese respondents reported having their blood pressure measured, and 36% of overweight and 50% of obese respondents reported having a test for diabetes.

Conclusion: These findings have implications for chronic disease identification and management.

Keywords: overweight, obesity, clinical practice guidelines, diabetes, hypertension, chronic disease

Introduction

Obesity rates in Canada, as in other parts of the developed world, have increased dramatically over the last few decades.[1,2] Since obesity is strongly linked to a number of chronic diseases, including diabetes, coronary heart disease and hypertension,[1] rates of these are on the increase. As a result, the sustainability of the Canadian health care system is significantly affected.[3] A recent analysis estimated the total direct costs attributable to overweight and obesity at $6.0 billion, which corresponds to 4.1% of total Canadian health care expenditures.[3]

Canadians with a body mass index (BMI) greater than 30 kg/m^2 are 4 times as likely to have diabetes, 3.3 times as likely to have high blood pressure and 1.5 times as likely to have heart disease.[1] A recent US study suggested that, given the marked increase in the proportion of obese people, obesity has become an equal if not greater contributor to the burden of disease than smoking.[4] Further, compared to baseline rates the prevalence of Class II (BMI 35–39.9 kg/m^2) and Class III obesity (BMI ≥ 40 kg/m^2) in Canada has shown the biggest increase.[5] In other words, those who are overweight or obese—the very individuals at greater risk of these comorbidities—are getting heavier faster.[4]

Yet, despite these stark statistics, obesity is not well managed within the current health system, a situation not unique to Canada.[6,7] Few health care professionals advise their patients about weight management in general or provide obesity management services.[8] Indeed, many Canadian health care professionals fail to even raise the issue of obesity with their patients.[9] Against this backdrop, publication of the 2006 Canadian Clinical Practice Guidelines for the management and prevention of obesity marked an important milestone in addressing obesity.[10] These Guidelines aim to effect change in clinical practice and ultimately decrease the prevalence of obesity and its complications among Canadians.[11] They make 72 recommendations to support obesity management across a range of settings, emphasizing the role of health care professionals within the Canadian health care system.

We know very little about the current weight management experiences of Canadians; nor do we know whether their experiences reflect the recommendations within the Guidelines. Understanding more about the weight management experiences of overweight and obese people could encourage them to seek appropriate

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weight management support. In addition, insight into these individuals' experiences in relation to the recommendations in the Guidelines may shed light on the weight management practices of health care professionals and help improve their application of the Guidelines. Given the large number of recommendations in the Guidelines, it is not feasible to measure them all within routine clinical practice. However, there are three that are particularly relevant to chronic disease identification and management.[10] These are No. 4, which recommends the measuring waist circumference in all adults to assess obesity-related health risks, and Nos. 6 and 7, which address the need to screen for obesity-related health risks and complications.[10] Reporting the weight management practices associated with these three recommendations by overweight and obese individuals therefore serves as a proxy measure of implementation of the Guidelines by health care professionals.

Methods

This survey sought to determine the weight management experiences of Canadian adults, that is, whether those who are overweight or obese (1) reported seeking support from family physicians or other health care professionals for weight management; and (2) reported weight management experiences that reflected three recommendations from the guidelines, namely measuring waist circumference (recommendation No. 4) and screening for weight-related comorbidities such as hypertension or diabetes (recommendations Nos. 6 and 7).

On behalf of the federally funded Canadian Obesity Network, Ipsos Reid conducted a general population omnibus survey over two consecutive weeks between March 23rd and April 3rd, 2009. Each one-week wave of the survey involved around 1000 adult Canadians. An independent panel of experts developed the interview questions, including 23 related to weight management practices as well as those related to demographic characteristics, including self-reported height and weight, gender, age, income and region of residence. Canadian SHIELD Ethics Review Board gave ethical approval. The sample size was chosen to provide robust data from a representative cross-section of the population based on key demographic criteria (e.g. sex, age, location).[12]

Interviews were conducted using a computer-aided telephone interview (CATI) system and random digit-dialing. Data were weighted to be nationally representative of all adults aged 18 years plus and balanced to match the most recent Statistics Canada figures for sex, age, income and region of residence.[12] The proportions and means were statistically compared within 5% significance level ($p < .05$). BMI was calculated based upon self-reported height and weight and respondents were categorized as underweight (< 18.5 kg/m^2); normal weight (18.5–24.9 kg/m^2); overweight (25–29.9 kg/m^2); and obese (≥ 30 kg/m^2). In addition, two further categories, BMI of 27 kg/m^2 plus with comorbidities and BMI of 30 kg/m^2 plus without comorbidities, were calculated. These categories represent established cut-offs as outlined in the Guidelines document.[10] As the focus of the survey was weight management experiences in relation to overweight and obesity, data on the small number of underweight respondents (n = 52) are not reported separately in this analysis.

Results

Sample characteristics

A total of 2004 survey respondents were included in the analysis. Of these, 48% were men (n = 970) and 52% women (n = 1034); 78% were interviewed in English (n = 1557) and 22% in French (n = 447). The mean age of respondents was 47 years (standard deviation of 16.4); 28% were younger adults (18–34 years; n = 559), 39% were middle-aged (35–54 years; n = 788) and 32% were older adults (≥ 55 years; n = 649). Seven respondents declined to give their age. Of the sample, 81% of were classified as urban dwellers (n = 1615), while 19% were rural dwellers (n = 388). The mean BMI was 26.5 kg/m^2 (27.3 kg/m^2 for men and 25.7 kg/m^2 for women), with 32% of respondents classified as overweight (n = 651) and 21% as obese (n = 411). Nevertheless, 44% described their weight as "about right" (n = 874) and 37% described themselves as slightly overweight (n = 734), with only 3% describing themselves as obese (n = 51). This pattern was generally consistent regardless of sex or age. Of those classified as overweight, 38% rated themselves as "about right" (n = 248/651); of those classified as obese, 4% rated themselves as "about right" (n = 18/411); and of those classified as normal weight, 73% correctly described their weight as "about right" (n = 567/779). Of those surveyed, 41% reported that they had never tried to lose weight (n = 818). More women (52%) than men (37%) had tried to lose weight in the previous 12 months; this difference increased with increasing age, with a similar pattern reported by those who tried to lose weight more than 12 months before the survey.

Objective 1: Weight management experiences of Canadians

We sought to determine whether overweight or obese Canadian adults reported seeking support from family physicians or other health care professionals for weight management. The majority of overweight (n = 418/651; 64%) and obese (n = 363/411; 88%) respondents had tried to lose weight, most during the previous 12 months, with similar rates for those with a BMI of 30 kg/m^2 plus without comorbidities (ever tried losing weight; n = 188/221; 85%) and BMI of 27 kg/m^2 plus with comorbidities (ever tried losing weight; n = 253/293; 86%). Differences in having tried to lose weight within the previous 12 months were significant for overweight compared with normal weight respondents, and for obese compared with normal weight and overweight respondents. Table 1 shows the characteristics of survey respondents' weight management experiences.

The majority of respondents (n = 1711; 85%) had seen a physician (for any condition, not specifically because of their weight) at least once in the 12 months before the survey. The proportions were generally similar among all groups, although some patterns were apparent; more women (90%) than men (80%), a greater percentage of the oldest age group (≥ 55 years; 91%) than the two younger age groups (18–34 years; 35–54 years;

TABLE 1
Characteristics of survey respondents' visits to a physician and weight loss attempts

Respondent categories	Sample size, n	Visits to physician			Weight loss attempts		
		Seen a physician in the past 12 months[a], n (%)	Mean no. of times seen a physician[b]	Under physician's care for any condition, n (%)	Last tried to lose weight in the past 12 months, n (%)	Last tried to lose weight over 12 months ago, n (%)	Never tried to lose weight, n (%)
All[c]	2004	1711 (85%)	2.86	808 (40%)	897 (45%)	283 (14%)	818 (41%)
Normal weight (BMI 18.5–24.9 kg/m²)	779	638 (82%)	2.58	247 (32%)	214 (27%)	100 (13%)	462[d,e] (59%)
Overweight (BMI 25.0–29.9 kg/m²)	651	559 (86%)	2.65	272[f] (42%)	326[f] (50%)	92 (14%)	230[e] (35%)
Obese (BMI ≥ 30.0 kg/m²)	411	364[f] (89%)	3.47	233[d,f] (57%)	290[d,f] (71%)	73[f] (18%)	41 (10%)
BMI ≥ 30.0 kg/m² with no comorbidities	221	178 (81%)	2.82	43 (19%)	139 (63%)	49 (22%)	32 (15%)
BMI ≥ 27.0 kg/m² with comorbidities	293	289 (98%)	3.96	293 (100%)	210 (71%)	43 (15%)	40 (14%)

Abbreviations: BMI, body mass index; n, sample size; *p*, p-value.

[a] Weighted data.

[b] Based on all respondents, including those who said that they had not seen a physician in the last 12 months. Median number of visits = 2.0.

[c] Data for underweight respondents (n = 52) or respondents who did not know or refused to provide their weight (n = 111) are included in these totals, but are not presented elsewhere in the table.

[d] Significantly different from overweight respondents ($p < .05$).

[e] Significantly different from obese respondents ($p < .05$).

[f] Significantly different from normal weight respondents ($p < .05$).

83% each), and an increase in the number of visits with increasing BMI. The highest consultation rate was seen in those with BMI of 27 kg/m² plus with comorbidities (n = 289/293; 98%). The mean number of visits was based on all respondents, including those who said that they had not seen a physician in the previous 12 months; as a result, the median number of visits, 2.0, was also calculated.

While 67% of all respondents reported that they had not specifically asked a health care professional about losing weight (n = 1339), of those that had (n = 665) 72% had consulted their family physician (n = 482), 24% consulted a dietitian (n = 158), 16% a nutritionist (n = 108), 16% a nurse or nurse practitioner (n = 107) and 9% a pharmacist (n = 59). Percentages do not add up to 100 as some respondents consulted more than one health care professional. Consultation rates were higher for women than men and increased across the age groups and with increasing BMI.

Only 15% of respondents had asked their family physician about weight loss (n = 295) in the 12 months before the survey; slightly more women (17%) than men (13%) sought such help. Again, there was a trend with increasing age, from 10% (18–24 years) to 18% (≥ 55 years), and BMI, with 24% and 56% of the overweight and obese respondents having ever asked a physician about losing weight. Of these, around half had done so in the 12 months before the survey (i.e. 13% and 35% overweight and obese respondents, respectively).

Forty percent of all respondents reported being currently under the care of a physician for some condition (n = 808/2004). Slightly more women (44%) than men (37%) reported being currently under the care of a physician for any condition, and this increased with age, from 19% in young adults (18–34 years) to 66% in older adults (≥ 55 years). Similarly, there was an age-related increase in the percentage under care for specified weight-related comorbidities, ranging from 6% to 56%. Around half of the respondents who were overweight (n = 272/651; 42%) or obese (n = 233/411; 57%) were under the care of a physician for any condition, with 30% and 46%, respectively, for specified weight-related comorbidities (n = 193 and n = 190 respectively). Table 2 shows the self-reported prevalence of the most common medical conditions.

It is interesting to note that only 30% of overweight or obese respondents had been advised to lose weight without specifically asking their physician (n = 320/1062). In 19% of overweight or obese respondents (n = 197/1062), this advice was understood to have been given to improve health in general, and in 12% (n = 123/1062), to improve the treatment of some other medical condition. This pattern was similar for men and for women. However, advice to lose weight was more often given to middle-aged (35–54 years; 21%) and older adults (≥ 55 years; 24%) than younger adults (18–34 years; 11%), and increased with increasing BMI (normal range, 5%; overweight, 18%; obese, 49%), with advice given most frequently to those with a BMI of 27 kg/m² plus with comorbidities (54%). Obese respondents understood that the advice to lose weight was meant to improve their overall health (30%) and to improve the treatment of some other medical condition (19%). Similarly, those with a BMI of 27 kg/m² plus with

TABLE 2
Self-reported prevalence of common medical conditions by BMI category

Medical condition	Respondents[a], n (%)			
	All (n = 2004)	Normal weight[b] (n = 779)	Overweight[c] (n = 651)	Obese[d] (n = 411)
High blood pressure	301 (15)	67 (9)	99 (15)[e]	111 (27)[e,f]
High cholesterol	211 (11)	48 (6)	78 (12)[e]	74 (18)[e,f]
Cardiovascular/heart disease	99 (5)	23 (3)	35 (5)[e]	33 (8)[e]
Diabetes	146 (7)	35 (5)	51 (8)[e]	55 (13)[e,f]
Osteoarthritis	134 (7)	36 (5)	43 (7)	51 (12)[e,f]
Other	369 (18)	134 (17)	116 (18)	88 (22)

Abbreviations: BMI, body mass index; n, sample size; *p*, p-value.

[a] Weighted data.

[b] BMI = 18.5–24.9 kg/m^2.

[c] BMI = 25.0–29.9 kg/m^2.

[d] BMI ≥ 30.0 kg/m^2.

[e] Significantly different from normal weight respondents ($p < .05$).

[f] Significantly different from overweight respondents ($p < .05$).

comorbidities understood that this advice had been given to improve health in general (28%) and to improve the treatment of some other medical condition (26%).

Of the 383 overweight or obese respondents who had requested a health care professional's support to lose weight, 15 (4%) reported receiving no advice, while 261 (68%) reported receiving dietary advice, 237 (62%) reported receiving exercise advice, 47 (12%) were advised to join a weight loss program, slimming club, or to take meal replacements or supplements, and 15 (4%) were prescribed anti-obesity medication. Of the 230 obese respondents who had requested support to lose weight, 10 (4%) reported being advised on options for surgery, as did one overweight individual.

Objective 2: Reported weight management practices of family physicians consistent with the Canadian Guidelines

We sought to determine whether survey respondents reported that their family physicians used weight management practices as recommended in the Guidelines.[10] Table 3 shows the number of respondents who reported having their waist circumference measured in the previous 12 months as well as their blood pressure and blood glucose tested. Only 14% of respondents reported that a physician had measured their waist circumference (n = 285/2004); this was slightly higher for men (17%) than women (12%) and increased with age from 11% in younger adults (18–34 years) to 14% and 18% in middle-aged (35–54 years) and older adults (≥ 55 years) respectively. Measuring waist circumference in the previous year was only slightly influenced by BMI (normal weight, 13%; overweight, 14%; obese, 18%) and only marginally higher in those with BMI of 27 kg/m^2 plus with comorbidities (21%) relative to the obese, although the difference between normal weight and obese respondents was significant. However, 84% of overweight or obese respondents had not had their waist circumference measured in the previous year (n = 889/1062).

TABLE 3
Reported weight management practices of family physicians using three of the 2006 Canadian Clinical Practice Guidelines

Respondent categories	Sample size, n	Clinical Practice Guideline[a]								
		Waist circumference, n (%)			Blood pressure, n (%)			Diabetes test, n (%)		
		In the past 12 months	Not in the past 12 months	Don't know	In the past 12 months	More than 12 months ago	Never	In the past 12 months	More than 12 months ago	Never
All	2004	285 (14)	1700 (85)	19 (1)	1603 (80)	313 (16)	80 (4)	723 (36)	443 (22)	812 (41)
Normal weight (BMI 18.5–24.9 kg/m^2)	779	100 (13)	672 (86)	7 (1)	581 (75)	150 (19)[b,c]	43 (5)[b,c]	221 (28)	166 (21)	382 (49)[b,c]
Overweight (25.0–29.9 kg/m^2)	651	92 (14)	554 (85)	6 (1)	536 (82)[d]	93 (14)	20 (3)[c]	235 (36)[d]	151 (23)	254 (39)
Obese (≥ 30.0 kg/m^2)	411	73 (18)[d]	335 (82)	3 (1)	358 (87)[d]	50 (12)	3 (1)	206 (50)[b,d]	98 (24)	104 (25)
BMI ≥ 30.0 kg/m^2 with no comorbidities	221	32 (14)	187 (85)	1 (0)	173 (78)	45 (20)	3 (1)	75 (34)	61 (28)	82 (37)
BMI ≥ 27.0 kg/m^2 with comorbidities	293	61 (21)	230 (79)	2 (1)	283 (97)	10 (3)	0 (0)	186 (63)	62 (21)	44 (15)

Abbreviations: BMI, body mass index; n, sample size; *p*, p-value.

[a] Weighted data.

[b] Significantly different from overweight respondents ($p < .05$).

[c] Significantly different from obese respondents ($p < .05$).

[d] Significantly different from normal weight respondents ($p < .05$).

With regard to measuring blood pressure in the previous 12 months, there was a trend with gender and age. Slightly more women (85%) than men (75%) reported that their blood pressure was checked, and 69% of younger adults (18–34 years) compared to 90% of older adults (≥ 55 years).The trend also increased with increasing BMI. Slightly more women (65%) than men (51%) reported ever having had a test for diabetes, and this also increased with age (40% in those aged 18–34 years up to 74% in those aged 55 years plus) and increased BMI (36% of the overweight and 50% of the obese) in the previous 12 months. Differences were significant for both overweight and obese respondents compared with those in the normal weight category for blood pressure and diabetes testing within the previous 12 months.

Discussion

This survey offers insight into the current weight management experiences of Canadians and highlights the weight management practices of their physicians in relation to three of the recommendations of the 2006 Clinical Practice Guidelines. Of the survey respondents, 53% reported being overweight (33%) or obese (20%), slightly lower than published Canadian data (overweight, 36%; obese, 23%),[1] possibly as a result of using self-reported data on height and weight (individuals typically underestimate self-reported weight and overestimate self-reported height.[11]) It is also interesting to note that 40% of overweight or obese respondents described themselves as "about right." This phenomenon has been found in other studies and may be due to the normalization of excessive weight gain as obesity rates rise.[14,15] This has implications for health care professionals who may need to raise awareness of the health risks of overweight or obesity in their patients before offering any weight management advice.

While over half the survey respondents reported being overweight or obese, surprisingly few had asked for or received weight loss advice from a health care professional. The majority of survey respondents reported visiting their physician in the previous 12 months, significantly more so if they were obese, and almost three-quarters of the overweight and obese Canadian adults surveyed had tried to lose weight, 58% in the previous 12 months. Nevertheless, only 21% of overweight individuals reported seeking help from their physician in the previous year, suggesting that the majority viewed weight loss as their own responsibility. Moreover, less than one-third of overweight/obese individuals had ever been advised to lose weight by a physician (without specifically asking), a further indication of the widely held societal view that obesity is an issue of personal responsibility rather than a medical problem.[15] As a result, most weight loss attempts reported here were likely to have been initiated by the individual rather than as a result of advice from a health care professional, with diet and exercise being the two most frequent methods used. Support was also sought from dietitians, nurses and pharmacists, highlighting the role of these different health care professionals in weight management.

Of those overweight or obese individuals who did ask their physician about weight loss, they generally received advice on diet and exercise, with only 4% of obese individuals reporting receiving advice on surgery options. This is especially noteworthy given the promising role of surgery as the leading effective long-term treatment option for people with severe obesity.[16,17] Although the criterion for considering surgery is a BMI of 40 kg/m^2 plus or 35 kg/m^2 plus with comorbidities, whereas our sample were classified as obese if they reported a BMI of 30 kg/m^2 and over, surgical interventions should be considered within a portfolio of options for obesity management, as outlined in the available Clinical Practice Guidelines.[10]

In Canada, there are considerable opportunities for physicians to screen for comorbidities and they do so routinely; 84% of overweight or obese Canadian adults had their blood pressure checked in the previous year, which is in line with the 2009 Recommendations of the Canadian Hypertension Education Program.[18] Similarly, according to the Canadian Diabetes Association 2008 Clinical Practice Guidelines, individuals aged 40 years plus should be screened for type 2 diabetes using a fasting plasma glucose test every three years, while those with additional risk factors for diabetes should be screened earlier and/or more frequently.[19] This survey found that 66% of overweight or obese Canadians reported ever being knowingly tested for diabetes. These findings are encouraging; however, there remains a need to address obesity earlier in the trajectory of weight gain to prevent the onset of chronic disease.[3] While respondents reported that their physicians were following the main recommendations in the screening of hypertension and diabetes, this did not apply to the recommendations for assessing obesity by measuring waist circumference.[10,18,19]

Given the scale of the obesity epidemic, our findings highlight that surprisingly few overweight or obese patients reported receiving advice about weight management. This suggests that the health care system is not providing adequate obesity management services.[9] This is in spite of obesity being increasingly prevalent[3] and increasingly recognized as a disease state in its own right.[8] One of the reasons that physicians may be more likely to address hypertension and/or diabetes than obesity could be because these are recognized as diseases, whereas obesity is still only considered a risk factor for disease.[20] While this view has been challenged by obesity experts,[21] obesity is not widely accepted as a disease, and this may constitute a barrier to improved management. Further, the prevalent societal view is that obesity is a condition caused by lack of willpower and that overweight and obese people are weak-willed, sloppy and lazy.[22] These views are frequently shared by health care professionals and may interfere with how they engage overweight and obese individuals in discussing weight management.[22] Addressing issues of bias and stigma associated with overweight and obesity may therefore improve how weight management advice is both offered and received within the health system.[22]

Limitations

Limitations to this study are the use of self-reported data; as previously outlined, self-reports are known to underestimate

the prevalence of obesity.[11] In addition, there was no objective measure of physician behaviour, although this survey does serve as a proxy measure by providing insight into the behaviour of health care professionals as reported by their patients. Respondents were not asked about their perceptions of the quality of their health care experiences or their interactions with health care providers, including whether they had experienced bias regarding their weight.[22] Despite these limitations, given that advice from a physician is a powerful motivator for weight loss,[23] the fact that overweight and obese individuals were not routinely encouraged to lose weight by their physicians is a cause for concern. Changing how health care professionals view and manage obesity is an area that has been poorly researched, as evidenced by the small number of studies included in a recent Cochrane systematic review on this topic.[24] However, the available evidence does support the role of physicians and other health care professionals in obesity management,[24] offering hope for the future management of this condition. Canadian physicians have expressed a need to spend more time with patients and to decrease the number of patients seen per hour.[25] They are also willing to refer patients to other health care professionals, such as dietitians, although they have reported concerns with timely access to these professionals and see a need for a less time-consuming referral process.[25] There is clearly a need for the current obesity guidelines to be more effectively implemented and evaluated, and for more resources to support implementation, particularly at the point of care.[26] Finally, and perhaps most importantly, there is a need to overcome some of the issues of bias and stigma held widely by society, including health care professionals,[27] and to recognize that obese people often present with a range of other issues that may hinder their ability to lose weight, including mental health issues, chronic pain and family or social barriers.[27] While not measured within this survey, these issues are known to influence weight management initiation and maintenance.[22,26] Health care professionals need to be aware of and supportive of these issues if we are to improve obesity management practices within the Canadian health care setting.

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The National Population Health Survey's assessment of depression risk factor associations: a simulation study assessing vulnerability to bias

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Abstract

Background: In Canada, the major source of longitudinal information on major depression epidemiology has been the National Population Health Survey (NPHS). However, the timing of NPHS interviews may raise concerns about the quality of its estimates. Specifically, the NPHS interview assesses major depressive episodes (MDE) in the year before an interview, whereas the interviews are conducted 2 years apart. The objective of this study was to determine whether this aspect of the NPHS can be expected to introduce bias into longitudinal estimates of risk factor associations.

Methods: A simulation model was used to represent the underlying epidemiology and the expected results of a study adopting the NPHS approach to assessment of MDE. The model was used to explore the extent of the resulting distortion of estimates across a range of underlying hazard ratios.

Results: The simulations indicated that the timing and coverage of depression interviews in the NPHS would not introduce substantial bias. The model suggested that incidence would be underestimated as a result of episodes being missed, but that this would not substantially distort estimates of association.

Conclusion: The timing of interviews in the NPHS is not expected to cause biased relative risk estimates. NPHS estimates may, of course, be influenced by other sources of bias.

Keywords: major depressive disorder, mood disorder, epidemiology, longitudinal studies, simulation, mathematical model

Background

Improved predictions of the risk of occurrence of major depressive episodes (MDE) would help target preventive efforts and support clinical management decisions. Epidemiological data are useful for determining risk, but the literature on longitudinal studies is limited. Most psychiatric epidemiological studies have been cross-sectional and have focused on prevalence rather than incidence. Prevalence is affected by the duration of illness and does not necessarily reflect risk.

Internationally, literature on incidence studies is beginning to emerge. Notable examples are the Netherlands Mental Health Survey and Incidence Study (NEMESIS),[1,2] and the Dunedin Birth Cohort.[3] A national source of longitudinal data in Canada is the National Population Health Survey (NPHS).[4] The NPHS includes a diagnostic instrument for past-year MDE, the Composite International Diagnostic Interview-Short Form for Major Depression (CIDI-SFMD).[5] However, the NPHS has certain design features that may call into question the validity of its longitudinal estimates. NPHS interviews occur every 2 years whereas the CIDI-SFMD interview covers the previous year; thus the diagnostic interview does not necessarily capture all episodes occurring between NPHS cycles. In addition, the CIDI-SFMD does not determine the timing of episodes beyond determining the presence of symptoms during the same 2-week period in the year preceding an interview.

Most studies that have used NPHS data have evaluated episode incidence as the proportion of persons without MDE in the year preceding an initial interview[6] or in several NPHS interview cycles[7] who then experience an MDE in the year preceding a subsequent interview. However, this cannot be precisely interpreted either as a 1-year or 2-year incidence proportion. Other studies have used proportional hazard models to evaluate incidence,[8] but the fundamental issue of the timing of interviews remains. As a measure of annual incidence in the year preceding a follow-up interview, the CIDI-SFMD may be non-specific (since some of the episodes that have their onset earlier than the year covered by the interview may be included in the numerator of an incidence proportion); if interpreted as a measure

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of a 2-year incidence proportion, the CIDI-SFMD may be insensitive (since some of the episodes that have an onset more than 1 year prior to the second interview may resolve prior to the year preceding the interview and therefore may not be recorded).[9] However, the actual impact of this aspect of the NPHS approach to measurement is unknown.

Had the NPHS included a reference standard measure fully assessing the course of MDE in its cohort, the impact of these design issues could be explored using real data. In the absence of such a measure, we sought to explore these issues using a simulation model designed to represent both the underlying epidemiology and the NPHS measurement strategy for MDE.

Method

To address the objectives of this study, we developed a discrete event simulation model using the software Arena version 10.[10] We set up the model so that the simulation clock would cover the same duration of follow-up as was available from the NPHS at that time, from 1994 to 2006. Entities in the model represented people, members of the NPHS cohort at their time of entry into the study and in a non-depressed state (at risk of incident episodes) at that time. The simulation model depicted the underlying epidemiology by representing incidence and recovery from episodes; it simulated the experience of each entity from their time of entry in 1994 until the occurrence of an episode of depression or for a maximum of 12 years of follow-up. The model did not attempt to represent mortality. There were three possible simulation paths for each entity: (1) they could complete 624 weeks (12 years) of simulated observation without an episode; (2) they could have an episode at a time when it would have been detected, in which case the episode was recorded as an incident case and the entity was then removed from the simulation; or (3) they could have an episode associated with an onset and recovery time that would have rendered it undetectable given the timing of the NPHS measurement strategy, in which case the entity was returned to the part of the model simulating incidence. Entities following the third pathway could then follow either of the available paths, experiencing recurrences (or not) which could then be detected (or not).

The general goal of this simulation study was to develop a representation of the epidemiology and the NPHS measurement strategy so that we could explore the extent of distortion introduced by the NPHS measurement strategy (see Figure 1). A first step was to represent recovery from MDE. This was important since longer episodes occurring in a 2-year period would be more likely to persist into the second year of this interval, potentially affecting the extent of introduced bias. The description of episode duration relied initially on some NPHS estimates [depicted (a) in Figure 1], specifically an ordinal logistic regression model describing self-reported episode durations in the NPHS cohort in relation to age (which was found to be the most important determinant of episode duration).[11] We used an equation representing the time-dependent pattern of recovery in different age groups in the model and calibrated its parameters to the NPHS estimates. Once recovery was depicted, the model could represent MDE incidence while also representing the possibility that the timing of onset and recovery from an episode might result in it not being identified. For example, in order to be detected at the 1996 interview, an episode had to include at least 2 weeks of sufficient depressive symptoms in the year preceding the interview. Ratios of incidence in respondents exposed or not exposed to risk factors have various strengths of association with MDE, as they would appear in the NPHS data.

The type of equation chosen to represent incidence of MDE (and recovery from MDE) was one that could depict incidence as diminishing over time spent free of depressive episodes, as is expected clinically and as has previously been observed in the NPHS data.[9] Equation 1 was used to calculate a linear function (LF) for each entity, using an attribute (here labelled with the non-specific term "covariate") assigned to that entity as a value of 0 or 1 and representing a risk factor exposure:

$$LF_{incidence} = \alpha + \beta_{covariate} * covariate + \beta_{\log t} * \log t$$

where t is the time in weeks. The time interval represented in the simulations were evaluated in 1-week intervals within the simulation. For the sake of simplicity, Equation 1 includes only a single covariate term, an indicator variable assuming a value of 0 or 1. Age was, however, depicted in most simulations as an attribute at five levels (12–18 years, 19–25 years, 26–45 years, 46–65 years and 66 years plus) using four separate indicator variables (such that the 12–18 years age group was the baseline category) since age is the main determinant of episode duration.[11] The linear function (LF) was transformed into a weekly risk (that changed with each passing week in the non-depressed state) using Equation 2:

$$\text{Weekly risk} = 1 - \exp[-\exp(LF)]$$

We used this weekly risk equation to simulate the risk of a new episode during each week of the simulation. This was represented in the simulation path by a loop, with passage around this loop corresponding to 1 week of simulation time. With each transit around the loop the time variable counted up by 1 week and the risk was recalculated. Recovery from an episode

FIGURE 1
Schematic diagram depicting the simulation approach used in the study

Abbreviations: MDE, major depressive episode; NPHS, National Population Health Survey.

was simulated using a similar approach. In order to explore the relationship of βi coefficients from Equation 1 to analogous NPHS estimates, it was necessary to identify a value for α and $\beta_{log\ t}$, also from Equation 1. This we did by fitting a grouped time proportional hazard model for 7029 NPHS respondents with complete data collection across all of the relevant NPHS cycles. We chose this subset because an important MDE risk factor, family history of MDE, was only evaluated in the 2004 NPHS. Table 1 compares the characteristics of this cohort with those of the entire NPHS cohort. The main difference was that the n = 7029 cohort was younger, probably due to attrition of elderly respondents from the original sample (e.g. due to death or institutionalization) over time. Table 2 shows the estimated hazard ratios from the proportional hazard model. The α value (−7.435) and $\beta_{log\ t}$ (−0.128) parameters from Equation 1 were calibrated, using an automated procedure in Arena® called OptQuest to identify parameter values that allowed the model output to resemble the NPHS estimates.

In reality, the NPHS interviews do not take place at a single point in time, but rather occur over the course of several months. For the sake of simplicity, this aspect of the data collection was not represented in the simulation model. Instead, each entity was generated at week 0 (baseline) and the subsequent interviews were represented as occurring at 104 weeks, 208 weeks, 312 weeks, 416 weeks, 520 weeks and 624 weeks. Tracking variables recorded the times of simulated onset and resolution of depressive episodes for each entity and tracked weeks elapsed in the simulation for each entity. The detection of episodes in the NPHS was assumed to have occurred if an entity had 2 or more weeks in the depressed state during the 52 weeks before a simulated assessment time. In this way, the model was able to represent the "under-detection" of episodes expected to occur in the NPHS and also the "over-detection" whereby episodes from previous years persisted into the year before an interview. The experience of a large number of entities was simulated each time a simulation was run, allowing evaluations of frequency distributions.

TABLE 1
Characteristics of the NPHS sample at baseline and of the respondents eligible for inclusion in the analysis

	NPHS sample at baseline (N = 13 175), %	Eligible sample (n = 7029), (%)
Sex		
Male	48.4	46.4
Female	51.6	53.6
Age (years)		
12–18	11.8	11.9
19–25	10.2	8.8
26–45	40.6	44.8
46–65	24.5	26.8
66+	13.0	7.6
Marital status		
Married/common-law	59.8	65.3
Single	28.0	25.3
Widowed/separated/divorced	12.2	9.4
Education		
High school graduation or less	47.5	42.4
Some post-secondary or higher	52.5	57.6
Income[a]		
Lowest	17.5	12.6
Low/mid/high	82.5	87.4
Injuries in the past 12 months[b]		
Yes	17.1	17.4
No	82.9	82.6
Chronic condition		
Yes	49.9	48.0
No	50.1	52.0
Smoking status		
Current	28.3	25.3
Former/never	71.7	74.7
Childhood stress or trauma		
Yes	47.7	48.0
No	52.3	52.0
Stress[c]		
Yes	28.5	27.8
No	71.5	72.2
Mastery[d]		
Low	25.5	23.5
Not low	74.5	76.5
Self esteem		
Low	34.6	32.8
Not low	65.4	67.2
Benzodiazepine use in the past 2 days		
Yes	2.0	1.4
No	98.0	98.6
Pain		
Moderate/severe	10.8	9.7
Mild/no	89.2	90.3

Abbreviation: NPHS, National Population Health Survey.

[a] The lowest income group corresponds to an income of less than $15,000 for a household of 1–2 persons, $20,000 for a family of 3–4 persons and less than $30,000 for 5 or more persons.

[b] Affirmative responses to the question: "In the past 12 months did you have any injuries serious enough to limit your normal activities?"

[c] Upper quartile scores on a scale containing up to 16 questions concerned with ongoing sources of stress.

[d] Mastery is the extent to which individuals believe that their life chances are under their control. This was assessed in the NPHS on a scale of 7 questions.

TABLE 2
Estimated hazard ratios from the NPHS

Variable	Hazard ratios estimated directly from the NPHS data
Female	1.5
Age group, years[a]	
19–25	1.0
26–45	0.8
46–65	0.5
66+	0.3
Injury	1.3
Chronic condition[b]	1.3
Current smoking	1.3
Childhood stress[c]	1.4
Stress[d]	1.5
Mastery[e]	1.3
Family history of depression	1.6
Pain[f]	1.8
Benzodiazepine use	1.8

Abbreviations: MDE, major depressive episodes; NPHS, National Population Health Survey.

[a] Baseline category is the 12–18 year age group.

[b] One or more reported conditions.

[c] Any one or more reported childhood traumas.

[d] Upper quartile on a stress scale. The scale contained up to 16 questions concerned with ongoing sources of stress.

[e] Lower quartile on a mastery scale. Mastery is the extent to which individuals believe that their life chances are under their control and was assessed in the NPHS on a scale consisting of 7 questions.

[f] Assessed using items from a scale associated with the Health Utility Index.

The model was initially verified by assessing expected outputs associated with various input values. For example, the frequency of missed episodes was evaluated in relation to various episode durations. When the recovery rate from episodes was represented as being very high, for example, by entering a large value for the α coefficient in the recovery equation (see Equation 1), the proportion of entities with undetected episodes became approximately 50%; when the recovery probability was very low (e.g. a large negative value for the same coefficient), the proportion of entities with undetected episodes became 0. To calibrate the model, we created output variables representing the sum of squared differences between NPHS episode duration estimates and used the simulated output and OptQuest to identify parameter values that allowed the model output to resemble the NPHS estimates. We also identified values for the recovery equation that produced simulated episode duration frequencies resembling those predicted by the ordinal logistic regression model describing age-specific episode durations in the NHPS.[11]

These approximate representations of episode duration were then used to explore the relationship between NPHS estimates of hazard ratios for risk factors and a series of hypothetical hazard ratios crossing a range of relevant values. The logarithm of this set of hypothetical hazard ratios was entered into the model as β coefficients in Equation 1, where the β coefficients are log hazard ratios. The output from the model (reflecting the simulated onset, resolution and measurement of episodes), in the form of risk ratios for the first 104-week risk interval, was then compared to these hazard ratios to see how closely they agreed.

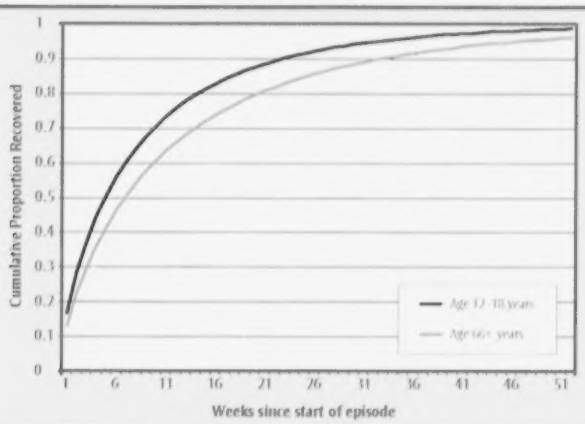
The NPHS used the CIDI-SFMD to assess MDE. This is a brief, fully structured interview designed to identify probable past-year episodes. The CIDI-SFMD interview is designed for use by non-clinician interviewers and is scored with a predictive probability algorithm based on the number of symptom-based criteria fulfilled during a 2-week period in the preceding year. Either depressed mood or loss of interest or pleasure, most of the time and nearly every day during that same 2-week period is required by the scoring algorithm, consistent with the Diagnostic and Statistical Manual of Mental Disorders (DSM-IV).[12] In data analyses for this study (as in most other NPHS-based depression studies), the instrument was scored at the 90% predictive cut-off point.[5] This cut-off requires endorsement of 5 of 9 specified depressive symptoms during the same 2-week period, a standard that is also broadly consistent with DSM-IV criteria. (While the approach taken by the CIDI-SFMD is consistent with DSM-IV, it should be noted that the instrument was developed using DSM-IIIR data collected in the National Comorbidity Survey.[5])

Results

Figure 2 shows durations of simulated episode for the youngest (12–18 years) and oldest (66+ years) age groups. These had the shortest and longest episodes, respectively. The curves for other age groups fell between these two. The shape of all the age-specific curves were broadly consistent with other international estimates.[11] As described above, these curves were then included in subsequent simulations as a representation of the recovery pattern and as a means of assessing the likely impact of the NPHS measurement strategy. Figures 3 and 4 show simulated predictions (200 000 simulated entities for each data point) of what the NPHS would be expected to identify as the relative risk given underlying hazard ratios of 1 to 5, according to the model. The logarithms of these various hazard ratios were used as β coefficients for incidence in producing these simulations, as in Equation 1. Two age groups are represented: one that includes those who were aged 12 to 18 years in 1994 (see Figure 3) and one that includes those who were 66 years plus in 1994 (see Figure 4). The hazard ratios in both age groups correspond very closely. The grey line on the figures represents equivalence of the two sets of hazard ratios. How we approach measuring MDE in the NPHS appears to make a negligible difference to the relative risk estimates arising from the approach.

Whereas our simulations indicate that hazard ratio estimates for MDE risk factors are not likely to be biased substantially

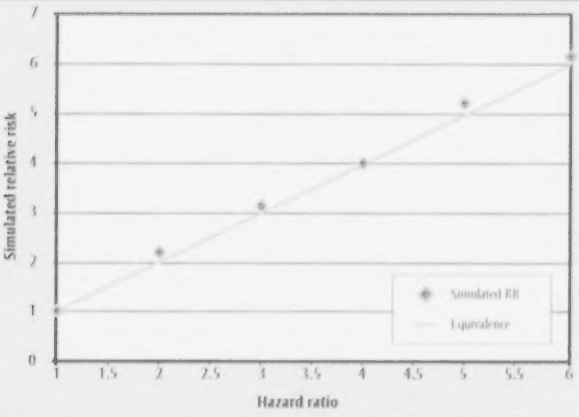
FIGURE 2
Pattern of recovery from MDE in the NPHS, in 2 age groups



Abbreviations: MDE, major depressive episode; NPHS, National Population Health Survey.

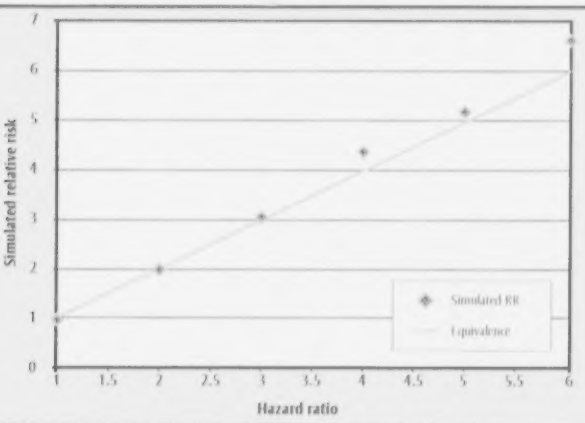
Note: The curves for the 19–25, 26–45 and 46–65 year age groups fall between those for the 12–18 and 66 plus year age groups. They are not shown to prevent clutter.

FIGURE 3
Simulated NPHS relative risks across a range of plausible hazard ratio values, 12–18 years



Abbreviations: NPHS, National Population Health Survey; RR, relative risk.

FIGURE 4
Simulated NPHS relative risks across a range of plausible hazard ratio values, 66+ years



Abbreviations: NPHS, National Population Health Survey; RR, relative risk.

by the timing of the NPHS interviews, incidence estimates are subject to bias. At strengths of association in the most relevant range (hazard ratios of 1 to 2), approximately 40% of entities with at least one MDE would not have been detected during follow-up, according to the simulations. Approximately 15% of the cases that would be regarded as incident (not depressed at one cycle, depressed at the next cycle) would have had their onset in the first year of the 2-year interval and could be considered false positives if the intention is to estimate a 1-year incidence proportion.

Conclusion

Psychiatric epidemiology is a fairly young discipline. The first of the current generation of studies (those employing standardized diagnostic procedures in representative samples) occurred in the 1980s.[14] To date, the literature has been largely descriptive and mostly cross-sectional. As a result, many estimates of prevalence are available, although these estimates have not been as consistent as might have been hoped.[15] A comprehensive understanding of the epidemiology of this condition will depend on longitudinal data clarifying the association of MDE incidence with various potential determinants, and associations between those determinants and the prognosis of MDE. Unfortunately, with the exception of a few international studies,[1,16] longitudinal data are scarce. In Canada, for example, the NPHS has been the major source of information on incidence[7] and of associations between longitudinal risk factors.[6] Unfortunately, aspects of the NPHS study design may cast some doubt on the validity of these estimates. In this sense, the results of this simulation study are encouraging because the simulations reported here do not suggest that hazard ratio estimates from the longitudinal NPHS data are likely to be substantially biased by the design features of the study.

Limitations and strengths

A notable limitation of our study involves the way in which incidence was depicted in the model. Rather than the incidence of depressive disorders, it was necessary to simulate MDE incidence. Some of those

considered "at risk" of MDE by virtue of not having had MDE at the baseline interview may actually have had major depressive disorders in the past and their incident episodes may have been recurrences of those disorders. The pattern of declining incidence over time represented by Equation 1 may be due partially to the gradual removal of those at highest risk from the population as they experience episodes.

Additional factors may, of course, affect the validity of estimates arising from the NPHS. The CIDI-SFMD is a brief diagnostic interview that does not include all of the detailed symptoms covered by the full CIDI. The CIDI-SFMD does not include probes for carefully delineating the duration and severity of each symptom or for distinguishing between organic and non-organic etiology of symptoms.[17,18] Misclassification bias arising from measurement error associated with the CIDI-SFMD (unrelated to the timing of its administration) may also distort estimates arising from the NPHS. Another important issue is that of attrition over time due to factors such as loss to follow-up, mortality and institutionalization; if such attrition depends on the outcome (MDE) in a way that differs with respect to risk factor exposures, bias may result. The estimates used in this study arose only from respondents with complete data collection, which is pertinent to the question of the validity and generalizability of the estimates. As the NPHS estimates used in the project arose from a subset of the longitudinal cohort (those providing complete data over seven cycles) the results may not be generalizable to the population. The simulation methods used here were intended to address one specific concern about the NPHS—the timing of its interviews in relation to its measurement of depression. The results can reassure us on this specific point.

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Dietary supplement use and iron, zinc and folate intake in pregnant women in London, Ontario

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Abstract

Introduction: We examined the dietary intake of iron, zinc and folate, estimated from both food and supplement sources, in 2019 pregnant women who participated in the Prenatal Health Project (PHP). The PHP recruited pregnant women from ultrasound clinics in London, Ontario, in the years 2002–2005.

Methods: Participants completed a telephone survey, which included a food frequency questionnaire and questions on dietary supplement use. Frequencies of use of dietary supplements were generated. Nutrient intake values were estimated from food and supplement sources, and summed to calculate total daily intake values.

Results: Most women took a multivitamin supplement, and many women took folic acid and iron supplements; however, one-fifth of the sample did not take any supplements providing any of the three micronutrients. Despite being of a higher socio-economic status overall, significant proportions of the cohort ranked below the recommended dietary allowance values for iron, zinc, and folate. This suggests there may be other barriers that impact dietary practices.

Conclusions: Further research is required on how to better promote supplement use and a healthy diet during pregnancy.

Keywords: iron, folate, zinc, dietary supplement, diet, nutrition, pregnancy

Introduction

Adequate amounts of nutrients during pregnancy are essential for maternal, fetal and child health. However, few population-based studies have examined dietary intake and use of dietary supplements among pregnant women in Canada. Of particular interest are iron, zinc and folate. Iron is integral to the structure and function of red blood cells, and its deficiency can result in anemia. Anemia and iron deficiency during pregnancy can cause pre-term birth and low birth weight.[1] In non-anemic mothers, iron supplementation may offer protection against low birth weight.[2] Iron is also involved in myelination, neurotransmitter function, various cellular and oxidative processes, energy production and thyroid hormone metabolism.[1] Iron deficiency has been implicated in neurological and cognitive disorders in the mother; these include major depressive disorder, recognized to have health consequences on both the mother and child.[3,4] The 2009 Health Canada guidelines recommend a daily supplement of 16 to 20 mg of iron during pregnancy to ensure adequate iron intake.[5]

Zinc is integral to DNA synthesis and necessary for the structure and function of regulatory, structural and enzymatic proteins as well as cell membranes. It is involved in neurological function and proper immune function.[1,6,7] Various studies have implicated zinc deficiency in pre-term and low birth weight, although routine supplementation is not recommended unless there is an identified deficiency.[8] Zinc deficiency is also implicated in depressive disorders.[1,4] Folate is involved in the metabolism of nucleic acids and amino acids and in neurological functioning. While inadequate folate is implicated in various birth defects and poor pregnancy outcomes, its role in neural tube defects has received the most attention. In various countries, including Canada, women of child-bearing age are advised to take supplements. Food fortification policies are in effect in response to the strong evidence of the importance of folic acid intake in the very early stages of pregnancy.[9,10] Like iron and zinc, folate deficiency is implicated in depressive disorders.[1,4]

Health Canada has set out a Recommended Dietary Allowance (RDA) for a number of nutrients. The RDA is defined as the "average daily dietary intake level that is sufficient

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to meet the nutrient requirements of nearly all (97 to 98 percent) healthy individuals in a particular life-stage and gender group."[11] For pregnant women, the RDA of iron is 27 mg/day, of zinc is 11 mg/day and of folate, as dietary folate equivalents (DFE), is 600 μg/day.[11] In addition to the RDA, the Society of Obstetricians and Gynaecologists of Canada recommends a daily folic acid supplement of at least 400 μg (with higher amounts indicated based on risk status).[9]

In this paper, we examine the reported daily dietary intake of iron, zinc and folate estimated from both the food and dietary supplements of 2019 pregnant women in London, Ontario, who participated in the Prenatal Health Project (PHP). We also generate and examine rates of supplement use among the 2019 women and investigate the types of supplements taken among the entire PHP cohort (n = 2357). We present the results of analyses that explore possible sociodemographic determinants of dietary intake.

Methods

Data for the Prenatal Health Project (PHP) were collected between 2002 and 2005 from pregnant women recruited at ultrasound clinics in London, Ontario. The PHP was designed to assess the psychosocial, nutritional, endocrine and infectious determinants of pre-term birth, and its methods have been discussed in more detail elsewhere.[12] Inclusion criteria were being aged 16 years or older, living in the Greater London Area, carrying a singleton pregnancy of between 10 and 22 weeks gestation, and speaking English. Women who met the inclusion criteria and who signed the consent form were eligible to participate. Those who were carrying a pregnancy with a known fetal anomaly were excluded.

The Ethics Review Board for Health Sciences Research Involving Human Subjects at The University of Western Ontario approved the study.

Trained interviewers collected dietary supplement intake data as part of the structured PHP telephone survey questionnaire. They asked respondents for the name, amount and frequency of consumption of all nutrient supplements currently used regularly. Nutrient amounts could be quantitatively estimated if the participant reported the brand and product name of a prenatal supplement or else the exact nutrient amounts. When brand or product information of prenatal supplements was missing (n = 930), we calculated the nutrient composition from the most commonly used prenatal multivitamin supplement, Centrum Materna. (Of the 643 women of the PHP core cohort who named a brand, most used Centrum Materna [n = 592], followed by Life Brand [n = 24], the composition of which is identical to that of Centrum Materna.) To generate average daily values, we assumed the standard dose of one tablet per day; if otherwise specified, we adjusted the intake values according to the reported frequency.

In contrast to prenatal multivitamin products, where the similar compositions justify assuming nutrient content, regular adult multivitamins on the market vary substantially. Thus, for those who reported taking such a supplement without specifying a brand name (n = 137), nutrient intake from supplements was declared missing. For the same reason, single-nutrient dietary supplements without specific amounts provided were also declared missing, with the exception of folic acid. Because there is less variation among folic acid supplements, a dose of 400 μg folic acid per day was assumed for those who did not specify their regular dose of folic acid supplement. This assumption is consistent with other studies that have measured folate supplement intake.[13] Based on the reported frequency of consumption, we calculated average daily intake values. A few respondents claimed to be taking a separate folic acid supplement that provided more than 1 mg (1000 μg) of folic acid per day; due to the possibility of inaccurate reporting, these were not quantified but were declared missing. Folate intake from supplement sources was converted to dietary folate equivalents (DFE) by multiplying by 1.7.[11]

Dietary intake from food was assessed with a food frequency questionnaire (FFQ). This was given to the study participants to complete before their scheduled telephone interview; they then reported their answers during the telephone interview. The FFQ is considered an acceptable method of assessing dietary intake in large surveys, including prenatal studies.[14] The major advantages of the FFQ, which make it more practical than dietary recalls or multiple food records, are the low respondent burden and the lower cost of data collection since it can be incorporated easily into the telephone interview itself. As there was no specific information on the dietary intake of pregnant Canadian women, we based the design of the FFQ on dietary data collected through 24-hour dietary recalls from 183 women who were breastfeeding at three months postpartum.[15] We compared the FFQ to one developed for a USA-based study of prenatal health[14] and subsequently added some more foods (e.g. broth). A pilot test of the FFQ for the PHP was conducted in London, and the instrument was validated with 3-day food records kept by 22 pregnant women. The correlation coefficients were as follows: folate 0.76 ($p < .001$), zinc 0.46 ($p < .05$) and iron 0.19 (not significant).

The FFQ assessed the usual frequency of consumption of 106 foods over the month prior to the interview date. Frequency of consumption of each item was categorized as never, once per day, 2 to 3 times per day, 4 or more times per day, once per week, 2 to 4 times per week, 5 to 6 times per week, or 1 to 3 times per month. CANDAT Nutrient Calculation System software[16] was used to convert responses to metric estimates of energy and nutrient intake per day, based on the 2006 Canadian Nutrient File.[17] Nutrient intake values from supplements were added to those from food to yield total daily dietary intakes.

Of 3656 women approached at ultrasound clinics to participate in the PHP, 2747 agreed to participate and 2421 (66%) completed the survey. Of these 2421 respondents, 38 were eliminated from the "core" longitudinal cohort due to perinatal data not being available or not being applicable (for such reasons as loss to follow-up, miscarriage, abortion or neonatal death). Additionally, 26 women had been recruited into the study twice, for two different pregnancies; for each of these participants, a single set of data was randomly excluded. This

yielded 2357 PHP participants in the core longitudinal cohort, with 2019 included in the intake analyses reported in this paper. Those included had completed the FFQ, reported an energy consumption amount within two standard deviations from the sample mean (as outside that plausible range is indicative of possible inaccurate reporting), had plausible FFQ-derived intake values for the nutrients of interest, and did not have any missing values for the nutrient supplement intake values for the nutrients of interest.

Statistical analyses

For the sample of 2019 eligible study participants, we calculated descriptive analyses of the estimated mean daily intake values of iron, zinc and folate from food, from supplements and from total dietary intake. To explore the contribution of supplement use in this regard, stratum-specific mean intakes for each micronutrient were also calculated, based on whether a supplement containing the micronutrient was being taken; Student's *t* tests were conducted to see if the differences between strata of supplement use were statistically significant. A correlation matrix between the total intakes of the three micronutrients was also generated to see whether intakes were linked. To assess possible predictors of diet, we explored associations between diet and four categorical sociodemographic variables: age, marital status, education and household income. To this end, we used ANOVA to explore associations between the four categorical sociodemographic variables and total dietary iron, zinc and folate intake separately. In addition, we ran χ^2 tests between each of the socio-demographic variables and dichotomized supplement use to assess any possible associations. Finally, we determined the frequencies of the types of dietary supplements taken by the full core PHP cohort (n = 2357). The statistical package SAS version 9.1 (SAS Institute Inc.)[18] was used to conduct data management and statistical analyses.

Results

Table 1 shows the characteristics of the eligible survey participants (n = 2019). Most women were aged between 22 and 34 years (with mean age of 30 years), were married, had completed college or university and had household incomes of between $30,000 and $79,999. Most women reported taking one or more dietary supplements; however, 29.6% did not receive any zinc from supplement sources, 28.4% did not receive any iron from supplement sources and 20.3% did not receive any folic acid from supplement sources. Approximately one-fifth of the sample did not take any regular or prenatal multivitamin products or single-nutrient supplements that contained any zinc, iron or folic acid.

Table 2 shows the descriptive analyses of each of the three micronutrients. Included are estimates of mean daily intake from food, from dietary supplements and from both sources together. Also indicated is the proportion ranking below the RDA, based on the total dietary intake estimates. Because of the inherent limitations of the

TABLE 1
Characteristics of Prenatal Health Project (PHP) dietary intake analysis participants (n = 2019)

Categorical variables	Participants	
	Number, n	Percentage, %
Age group, years		
< 22	85	4.2
22–34	1578	78.2
35+	355	17.6
Marital status		
Married	1544	76.5
Common-law	310	15.4
Single/separated/divorced	163	8.1
Education[a]		
Completed college diploma/university degree	1431	71.3
Other	575	28.7
Household income[a], $		
< 30,000	224	11.9
30,000–79,999	941	50.0
80,000+	716	38.1
Using one or more dietary supplement(s)[b]		
Yes	1613	79.9
No	406	20.1
Taking a supplement containing iron		
Yes	1446	71.6
No	573	28.4
Taking a supplement containing zinc		
Yes	1422	70.4
No	597	29.6
Taking a supplement containing folic acid		
Yes	1610	79.7
No	409	20.3
Measured variables	**Mean**	**(SD)**
Age, years	30.4	(5.0)
Energy consumption, kcal/day	1982	(545)

Abbreviation: SD, standard deviation.

[a] Sample size is less than 2019 due to missing values.

[b] Containing any amount of folic acid, iron or zinc; therefore, those assigned a "no" to this variable were not taking any of the multivitamin products or single-nutrient products listed in Table 4.

TABLE 2
Intake from food and dietary supplements of iron, zinc and folate by pregnant women (n = 2019)

Micronutrient	RDA for pregnant women	Estimated mean daily intake, weight per day (SD)			Proportion of sample below RDA, %
		From food alone	From dietary supplements alone	Total	
Iron (mg/day)	27	13 (4)	19 (12)	32 (13)	31
Zinc (mg/day)	11	10 (3)	5 (3)	16 (5)	18
Folate (µg/day DFE)	600	473 (155)	1338[a] (763)	1811 (772)	16

Abbreviations: DFE, Dietary Folate Equivalent; RDA, Recommended Dietary Allowance; SD, standard deviation.

[a] In addition to the RDA from food, the Society of Obstetricians and Gynaecologists of Canada recommends that pregnant women take a 400 µg folic acid supplement (400 µg folic acid = approximately 680 DFE).[9]

FFQ method of dietary assessment, it is not considered appropriate to use the estimates of nutrient intake to assess nutrient adequacy. However, FFQ estimates may be used to rank nutrient intakes in a population based on the RDA.[19] In this case, a relatively high proportion of the sample fell below the specified RDA for all three micronutrients: iron (31%), zinc (18%) and folate (16%). A correlation matrix between the total intakes of the three micronutrients showed high correlation.

Table 3 shows stratum-specific mean estimates of total dietary intake, according to whether a supplement containing the particular micronutrient was being taken. The corresponding Student's *t* tests indicate statistically significant differences in mean intakes for all three micronutrients. Figure 1 shows histograms depicting the distributions of the three micronutrients. While total zinc intake follows a reasonably normal distribution, the distributions for total iron intake and total folate (DFE) intake are bimodal, each showing two distinct peaks. For both micronutrients, one peak was below the RDA level while the other was above. The peaks correspond to the stratum-specific mean estimates in Table 3; in other words, the bimodal distributions are a function of dietary supplement use.

Table 4 shows the types of nutrient supplement product used; that is, multivitamin products as well as single-nutrient supplements featuring iron, zinc or folate. To show the complete range of products used, the numbers are based on the full PHP core cohort of 2357 women. Therefore, the table includes entries that could not be quantified. Of the specified prenatal multivitamin supplements, the most commonly used product was Centrum Materna (n = 592). In the case of supplements of specific micronutrients (i.e. single-nutrient supplements, or products containing a small complex of nutrients), the most common were folic acid supplements (n = 354), followed by iron supplements (n = 98).

ANOVA tests were run to examine whether there were associations between the four categorical sociodemographic variables and each of total dietary iron, zinc and folate intakes. None was statistically significant. Similarly, of the χ^2 tests conducted between each of the sociodemographic variables and dichotomized dietary supplement use, none was statistically significant. In other words, age, marital status, education, and household income were not associated with either total dietary intake or supplement use in this group of women.

Discussion

London is a city in southwestern Ontario. In 2006, its population was just over 350 000.[20] The reported median family income in 2005 was $67,018—only slightly higher than the median family income for Canada ($63,866) and only slightly lower than that for Ontario ($69,156).[20] The results of this study may thus be informative for other Canadian cities with similar characteristics.

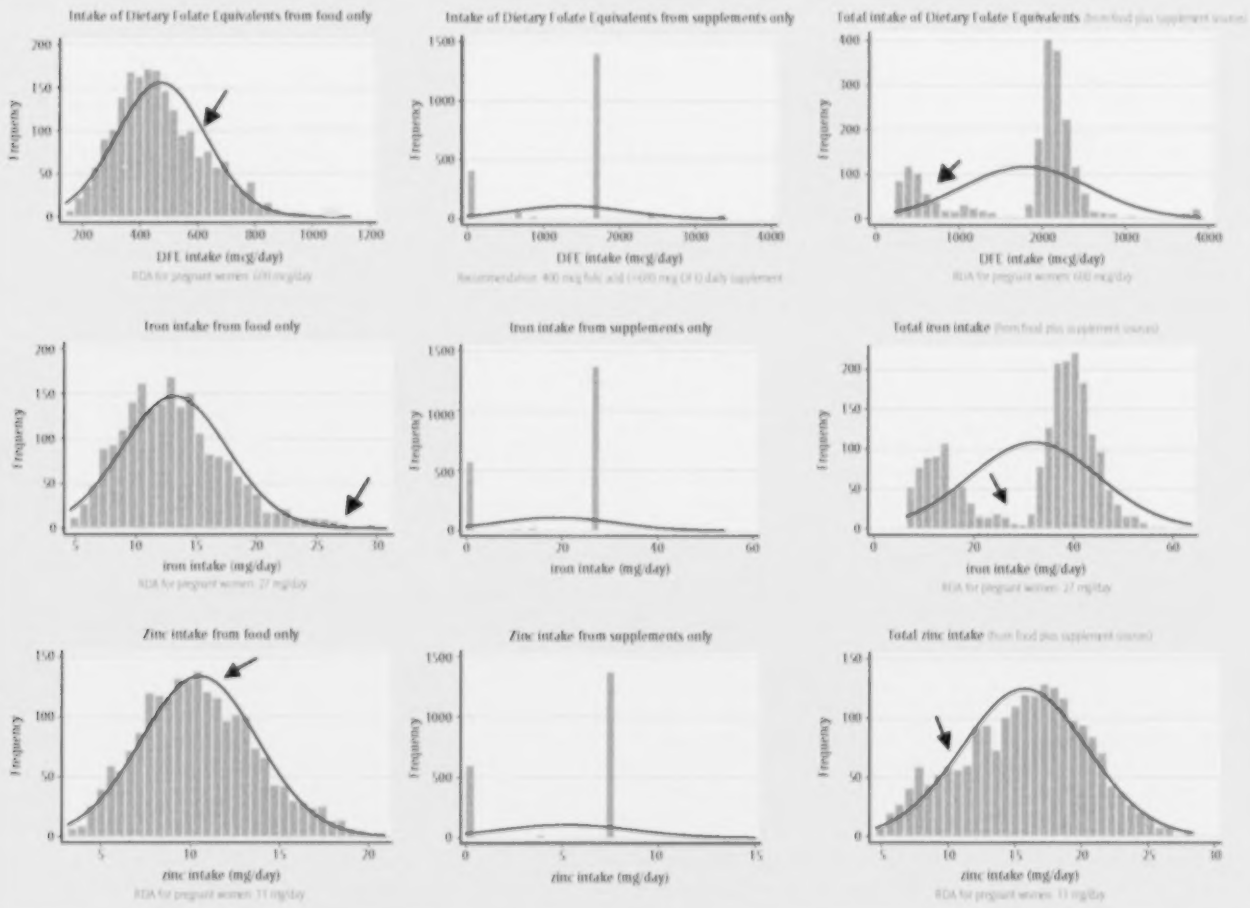
TABLE 3
Total dietary iron, zinc and folate intake by pregnant women, stratified by supplement use (n = 2019)

Micronutrient	RDA for pregnant women	Estimated mean daily intake, weight per day (SD) [n]		Student's *t* test[a]
		Participants obtaining micronutrient only from food	Participants obtaining micronutrient from both food and supplement sources	
Iron (mg/day)	27	13 (4)	40 (6)	−100.0
		[n = 573]	[n = 1446]	
Zinc (mg/day)	11	11 (3)	18 (3)	−44.7
		[n = 597]	[n = 1422]	
Folate (µg/day DFE)	600	482 (157)	2148 (422)	−78.4
		[n = 409]	[n = 1610]	

Abbreviations: DFE, Dietary Folate Equivalent; *p*, p-value; SD, Standard deviation.

[a] $p < .001$

FIGURE 1
Estimated dietary intake of dietary folate equivalents, iron and zinc from both food and supplement sources (separately and totalled) by Prenatal Health Project (PHP) survey participants (n = 2019)



Abbreviations: DFE, Dietary Folate Equivalent; RDA, Recommended Dietary Allowance.

Note: The arrows approximately indicate the RDA for each nutrient.

Few Canadian studies have examined dietary intake and supplement use during pregnancy. Of those that have, most focus on folic acid supplement use.[21-25] In terms of nutrient intake from food among Canadian pregnant women, a 2005 paper by Pick et al.[26] reported on the nutrient intake levels of a small sample of non-pregnant and pregnant women in Edmonton, Alberta. Because they were reporting findings from a pilot study, the sample size was relatively small (n = 52 pregnant women), which they acknowledge as a limitation.[26] In contrast, our study uses a very large sample size and a pre-piloted, validated instrument to capture dietary intake. Additionally, Pick et al. did not factor in nutrient values from dietary supplements,[26] while we were able to incorporate nutrient values from supplements to generate total intake estimates. Thus, our study offers a valuable glimpse into the nutritional status of a pregnant Canadian population, thereby contributing meaningfully to the literature in the area.

We found that a significant proportion of women had dietary intakes of iron, zinc and folate that ranked below the RDA values. Nutrient intake from food alone was particularly low (see Figure 1), supporting other Canadian studies that suggested that it is difficult for pregnant women to meet recommendations for key micronutrients from food alone.[26,27] Additionally, one-fifth of women did not take any supplements containing any of the three micronutrients. Given the importance of these micronutrients for maternal and fetal health, this is of concern.

Clinical practice guidelines emphasize the importance of folic acid supplementation during pregnancy[9] and recommend iron supplementation.[5] Thus it is not surprising that these two micronutrients were the most common among single-micronutrient products. The bimodal distributions associated with both micronutrients are a function of dietary supplement use, as shown in Table 3; women who used supplements for these nutrients were well above the RDAs for them and constituted

the higher-valued peaks, whereas women who did not use dietary supplements did not achieve the RDAs for them and constituted the lower-valued peaks. Thus, dietary supplement use is clearly integral to the attainment of the micronutrient intake levels required during pregnancy.

Even with folic acid fortification of foods in Canada and the United States, most women appear to require a separate folic acid supplement in order to achieve the red blood folate concentrations required to prevent neural tube defects.[25,27] While the exact mechanism by which folic acid prevents neural tube defects is unknown, it seems that folic acid supplements (rather than natural food folates) may be key to the preventive effect.[25] As such, it is of concern that 20% of women in this sample were not taking a dietary supplement containing folic acid.

As mentioned in the Results section, the intake values for the three micronutrients were highly correlated overall. This finding is likely a reflection of the fact that individuals tend to be deficient in multiple micronutrients due to poor overall dietary practices; along the same vein, it may also be a reflection of multivitamin supplement use, through which individuals receive the micronutrients together as a "package."

Age, marital status, education and household income were not associated with either total dietary intake of micronutrients or supplement use. As part of a separate analysis involving PHP data, we used multivariable regression to evaluate predictors of dietary zinc intake more thoroughly. Those findings, done in the context of a research question on the predictors of prenatal depression, have been reported in detail elsewhere;[28] none of the sociodemographic variables or psychosocial stress were shown to be predictors of dietary zinc intake in this cohort.[28] The cohort as a whole is of higher socio-economic status than the general population of the city of London.[30,28] Thus, other factors may account for the variation in dietary intake. Further investigation to uncover these factors would be pertinent from a public-health policy perspective. Certainly, the link between socio-economic status and dietary intake is well-established.[29-33] In that light, it is somewhat intriguing that notable proportions of a more socially advantaged cohort also show indications of inadequate dietary intake and a lack of supplement use. Such findings may flag the existence of additional barriers in Canadian women's lives, not captured by typical socio-economic status indicators. It has been suggested in the folic acid supplement literature, for example, that there may be barriers at the health care provider and public-health policy levels.[34,35] There has been increasing focus on the social determinants of population health and on health promotion as a function of public health;[34,36] both of these frameworks may be useful to understand the determinants of dietary intake and supplement use among Canadian women of childbearing age. Further research and action is warranted to help effectively promote healthy dietary practices across all segments of the Canadian population.

TABLE 4
Self-reported multivitamin supplements and single-nutrient supplements featuring iron, zinc or folate, taken by the full cohort of Prenatal Health Project (PHP) participants (n = 2357 women)

Source	Number of self-reported entries, n		
	Total	Quantified	Missing
Regular multivitamin supplement			
Product specified[a]	37	37	0
Product not specified	137	0	137[b]
Prenatal multivitamin supplement			
Product specified: Centrum Materna	592	592	0
Product specified: other[c]	51	51	0
Product not specified	930	930[d]	0
Iron			
Single-nutrient iron supplement	95	31	64[e]
Iron in a supplement with one other micronutrient	3	2	1[e]
Zinc			
Single-nutrient zinc supplement	2	2	0
Zinc with selected (few) other micronutrients	1	0	1[e]
Folate			
Single-nutrient folic acid supplement	347	315	32
Folic acid in a supplement with a few selected other micronutrients	7	3	4

Note: This table shows the frequencies of self-reporting of types of supplements. To show the full range of products used, the table is based on the core PHP cohort, including those participants who were excluded from the other analyses in this paper. Please note that some women may have been taking multiple types of supplements; as such, there may be multiple entries for a single participant. Similarly, as discussed in the paper and displayed in Table 1, a notable proportion of women did not take any supplements; there are no entries for these participants.

[a] Specified regular multivitamin brands: Centrum (regular), Centrum Forte, Centrum Protegra, Flintstones (children's multivitamins), Nutrilite Double X, Life Daily One for Women, Life Spectrum, Life Spectrum Forte, One A Day, One A Day – Women's.

[b] Declared missing because, in contrast to prenatal multivitamins, the nutrient composition of regular adult multivitamins varies substantially and therefore cannot be inferred from other brands.

[c] Specified prenatal multivitamin brands (apart from Centrum Materna): Equate, Fem, GNC, Jamieson, Life, Natural Factors (MultiStart), Orifer F, PregVit, Rexall, Thorne Research, Truly.

[d] Assumed to be identical to Materna since nutrient compositions of different prenatal multivitamin products are very similar.

[e] Declared missing because nutrient composition of single-nutrient dietary supplements (aside from folic acid) varies substantially and therefore cannot be inferred from other brands.

Study strengths and limitations

The FFQ method of assessing dietary intake offers only an estimate of nutrient values, and individual-level adequacy status cannot be determined with certainty.

However, for large survey studies such as this one, it is an acceptable and useful method, and still offers insight relevant for the purposes of public health. As noted in the Methods section, the correlation coefficient for the validation of iron intake is low (0.19). Since the estimate of iron intake from FFQ data was lower than that obtained from the 3-day records in the pilot test, our findings are likely conservative. However, this issue is unlikely to account for the very stark difference in total iron intake between those taking and not taking an iron-containing supplement. In other words, it likely does not alter the conclusion that supplement use is important in achieving the RDA for iron during pregnancy.

As described in the Methods section, we assumed that unspecified prenatal dietary supplements were of the same composition as Centrum Materna. This assumption, however, is reasonable given the similarities between popular prenatal multivitamin products on the market. Additionally, for those who did not specify the amount of their folic acid supplement, we assumed a daily dose of 400 µg of folic acid, though folic acid supplements do come in higher doses. Other studies that measured folic acid supplementation used a similar approach. The difference in mean DFE estimates is quite stark between those taking and those not taking folic acid supplements; as such, potentially underestimating folic acid intake for some participants as a result of this assumption would not alter our conclusions about the importance of supplementing with folic acid to achieve the RDA for folate.

The strengths of this study include the large sample size, the community-based sample, and the comprehensive assessment of both food and supplement sources in estimating dietary intake. Weaknesses of the study are the potential for selection bias and response bias. These are inevitable to some degree, although steps were taken to minimize their occurrence. Response bias was minimized by having trained telephone interviewers guide participants through all components of the questionnaire, including the FFQ. Selection bias was minimized by recruiting from ultrasound clinics in a variety of locations in London, where it is routine practice for pregnant women to get ultrasounds. A potential limitation is the fact that only English-speaking women could be recruited. However, as less than 2% of women in London, Ontario, cannot speak English,[10] the impact is likely negligible. The PHP cohort is of somewhat higher socio-economic status than the general population of the city of London, Ontario,[10] which may indicate selection bias; however, it also allowed for intriguing findings to come to light regarding dietary intake and supplement use in more socially advantaged segments of the population, as discussed above.

Conclusions

While the general importance of prenatal multivitamin supplement use is recognized in clinical practice guidelines,[9] zinc is not highlighted as a specific micronutrient of interest. While folic acid and iron supplementation are formally recommended, the data from this study suggest that, at the population level, dietary adequacy and supplement use may still be a concern. Notable proportions of this cohort showed lower dietary intake levels for all three micronutrients, as well as a lack of supplement use, despite being of a higher socio-economic status overall. Further research is warranted to evaluate both the comprehensiveness and the success of population-level implementation of current supplementation recommendations during pregnancy.

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National Epidemiologic Database for the Study of Autism in Canada (NEDSAC)

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In 2001, the Autism Spectrum Disorders—Canadian-American Research Consortium (ASD-CARC) launched a program of research on autism spectrum disorders (ASD). As part of that undertaking, and in response to concerns about the growing proportion of children diagnosed with ASD,[1,2] the National Epidemiologic Database for the Study of Autism in Canada (NEDSAC; www.nedsac.ca) was created as a multi-site ASD surveillance program. Government departments, clinicians and researchers* collaborated to establish regional teams to collect information (see Table 1) on children with ASD in British Columbia, Calgary (Alberta), Manitoba, southeastern Ontario,† Prince Edward Island, and Newfoundland and Labrador. NEDSAC provides estimates of the prevalence of ASD in Canadian children and a profile of those who are affected,[3] and allows researchers to monitor trends in age at diagnosis.[4] These data can help the health, education and social services sectors with planning and allocation of resources.[5,6]

TABLE 1
Data collected in NEDSAC

Demographics of children with ASD
• Date of birth and sex
• Number of biological siblings
• Number of biological siblings with confirmed or suspected ASD
• Mother's place of residence during pregnancy
• Parental ages at birth of child
• Ethnocultural identity
Diagnosis
• Type of professional(s) who made the diagnosis
• Tests or tools that were used
• Diagnostic subgroup
• Date of diagnosis
• Child's place of residence when diagnosed

Abbreviations: ASD, autism spectrum disorders; NEDSAC, National Epidemiologic Database for the Study of Autism in Canada

Estimating the prevalence of ASD

Different approaches are used to estimate the prevalence of ASD. One is to conduct population screening to identify suspected or diagnosed cases, followed by an assessment process to confirm or rule out the diagnosis. This approach is exemplified by a study done in Karlstad, Sweden: investigators used a combination of procedures to screen 826 children born there in 1985 and still living there in 1992.[7] The children were observed in

* A list of researchers and collaborators can be viewed at www.nedsac.ca under "Who we Are."

† Includes the six counties of Hastings, Prince Edward, Lennox & Addington, Frontenac, Leeds & Grenville and Lanark.

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various settings, and their parents and teachers were interviewed to assign a diagnosis. This approach has the potential to capture undiagnosed cases and allows for direct assessment to verify case status. However, low response rates, with potentially biased estimates, are a concern.[8] Moreover, it is too costly to use such an approach for ongoing surveillance in large populations.

A records-review approach is used by the Autism and Developmental Disabilities Monitoring Network in the United States.[9,10] Using standardized procedures and a common case definition, data are abstracted from health and education[1] records and reviewed by clinicians to assign case status.[9,10] This surveillance approach was deemed unfeasible in Canada: privacy legislation makes access to school records difficult, if not impossible, for researchers, and identifying cases through health sources alone could miss substantial numbers of children with ASD.[9,10]

A third approach relies on survey data to estimate the prevalence of ASD. For example, investigators in the United States analyzed responses to the 2007 National Survey of Children's Health; a child was considered to have ASD if the caregiver reported that the child had been diagnosed by a physician or other health care provider and still had the diagnosis.[11] Statistics Canada's quinquennial Participation and Activity Limitation Survey (PALS) contains a question specific to the occurrence of autism.[12] The PALS is not, however, a general population survey; its target population comprises individuals who responded "yes" to either of two questions on the Census concerning limitations of activity. Moreover, the Federal Government recently announced that it will no longer be conducting the PALS (Statistics Canada, personal communication, 30 June 2011).

A question on whether a health professional has ever diagnosed the child with autism was added to Cycle 8 (2008–2009) of the biennial National Longitudinal Survey of Children and Youth (NLSCY).[13] Statistics Canada cautions that the NLSCY is a general-population survey and is not designed for the analysis of relatively rare subpopulations, which could yield small samples and high sampling error. The initial sample for Cycle 8 of the NLSCY consisted of 35 795 children aged 0 to 7 years and youth aged 14 to 25 years. Cross-sectional weights, which are used to make inferences at the population level for the survey time period, are only available for the 0- to 7-year age group. It is unclear, therefore, how valid these survey data would be for estimating the prevalence of autism in Canada, particularly if they are stratified by age group and region.

Using administrative data is another option for surveillance, although these data demonstrate imperfect sensitivity and specificity.[14] This approach is only feasible in areas where the appropriate datasets can be linked for research purposes. In 2001, few provinces and territories had an administrative dataset infrastructure that could be used for ASD surveillance. (One notable exception was Manitoba: the Manitoba Centre for Health Policy houses health and education datasets that can be linked to estimate ASD prevalence. See the section at the end of this paper titled "NEDSAC now and in the future.")

For the reasons described above, none of the aforementioned approaches was considered feasible for ASD surveillance in Canada when NEDSAC was established. Instead, we formed partnerships with agencies that provide services to children with ASD in order to identify cases in the most cost-effective manner possible. This approach, similar to the one that uses administrative datasets to estimate prevalence, is apt to result in under-detection of cases for a number of reasons. First, the methods used by agencies to identify cases may mean that some children with ASD are missed. For example, in many education databases only one special education code is assigned in a given year, and so a child with ASD who also falls under another special education category may be coded under that category instead.[15] Service-based data also fail to capture children who meet research criteria for ASD but who have not been diagnosed. Across all Autism and Developmental Disabilities Monitoring Network sites, the proportion of children with a previously documented classification of ASD was lower than prevalence estimates obtained using the Network's surveillance methodology and case definition.[9,10] Similarly, findings from a population-based study in Olmsted County, Minnesota, revealed that only 46.8% of children who met research criteria for having ASD had a previous diagnosis.[16] Thus, the findings from NEDSAC should be interpreted as minimum prevalence estimates.

In contrast to relying on administrative datasets to estimate prevalence, however, identifying children with ASD through agencies that provide services to this population provides greater assurance that cases are "true positives" in the sense of having received a clinical diagnosis of ASD. It also enables the research team to contact families in order to directly assess and confirm the diagnosis in a subset of individuals.

Identifying children with ASD for NEDSAC

A considerable challenge with our surveillance approach is the use of provincial services such as health, education and publicly funded intensive behavioural intervention programs to identify cases. The delivery of these services, the ease of accessing data on children with ASD (i.e. whether information on these children is readily retrievable through service providers' databases or whether they have to review files to identify cases), and internal policies regarding data sharing vary widely across provinces and service providers. Accordingly, it proved impossible to use the same case ascertainment and data collection method in every region. Instead, regional protocols were designed to capture diagnosed cases of ASD in the most efficient way possible and to meet the information needs of agencies that provide the data (the latter explains the different age cut-offs among regions, as described below). Surveillance

[1] In 10 of 14 surveillance sites in 2002[9] and 6 of 11 surveillance sites in 2006[10].

was rolled out in 2002 and 2003 in six regions, and it continues in Manitoba, southeastern Ontario, Prince Edward Island, and Newfoundland and Labrador. Data collection was terminated in Calgary in 2006 and British Columbia in 2007. In the following sections, we describe the case ascertainment and data collection protocols in the surveillance regions. Table 2 summarizes these for the four regions where surveillance is ongoing.

In Prince Edward Island, the Department of Education and Early Childhood Development provides minimal data[5] to NEDSAC on preschoolers and school children aged less than 18 years diagnosed with ASD, including those who are home-schooled or attend private school. In Newfoundland and Labrador, the Department of Education and the Department of Health and Community Services provide minimal data[6] to NEDSAC on children aged less than 15 years diagnosed with ASD.[7] Children who are home-schooled or attend private school are not captured unless they are identified through the regional diagnostic teams. In

TABLE 2
Agencies in Prince Edward Island, Newfoundland and Labrador, southeastern Ontario and Manitoba that identify children with an autism spectrum disorder (ASD) for the National Epidemiologic Database for the Study of Autism in Canada

Region	Agency	Population served	Case ascertainment and data collection
Prince Edward Island	Department of Education and Early Childhood Development	Pre-school and school-aged children (includes those who are home schooled or attend private school)	The Department of Education and Early Childhood Development provides the research team with the date of birth, sex, prevalence year[a] and diagnostic subgroup for children < 18 years with an ASD, and sends information letters and consent forms to the parents or legal guardians. If a signed consent form is sent back to the research team, more detailed demographic and diagnostic information is collected by telephone interview with the parent or legal guardian.
Newfoundland and Labrador	Department of Education	School-aged children (excludes those who are home schooled or attend private school)	Similar to above, except limited to children < 15 years
	Department of Health and Community Services (Regional diagnostic teams operating in the four Regional Health Authorities, Intervention Services)	Collectively, all ages	
Southeastern Ontario[b]	Limestone District School Board	School-aged children (includes those who are home schooled)	Same as for Newfoundland and Labrador
	Upper Canada District School Board		
	Hastings & Prince Edward District School Board		
	Algonquin & Lakeshore Catholic District School Board		
	Catholic District School Board of Eastern Ontario		
	Conseil des écoles publiques de l'Est de l'Ontario		
	Conseil des écoles catholiques de langue française du Centre-Est		
	Child Development Centre, Hotel Dieu Hospital[c]	Children < 18 years	
	Pathways for Children & Youth, Autism Intervention Program	Children < 18 years who have been diagnosed with autistic disorder or considered to be on the moderate to severe end of the autism spectrum	
Manitoba	Children's Special Services, Manitoba Department of Family Services and Consumer Affairs	Children < 18 years, with the exception of those living on reserves	Agency staff review files and complete data collection forms for children < 18 years with ASD

Abbreviations: ASD, autism spectrum disorder.

[a] Year in which the child was first known to have an ASD diagnosis and reside in the surveillance region.

[b] Includes the six counties of Hastings, Prince Edward, Lennox & Addington, Frontenac, Leeds & Grenville and Lanark.

[c] Referral and assessment centre for children in southeastern Ontario with suspected developmental problems.

[5] Date of birth, sex, prevalence year (year in which the child was first known to have an ASD diagnosis and reside in the surveillance region), diagnostic subgroup.

[6] At the time of initial contact with the family or at the time of diagnosis, the regional diagnostic teams in Newfoundland and Labrador advise families of the NEDSAC study and the types of data provided to the researchers. In 2009, school principals in Newfoundland and Labrador sent a letter to parents and legal guardians of all school age children with ASD describing the NEDSAC project and the data that are collected. Parents and legal guardians were informed that they could contact the Department of Education directly to opt out of having any information on their child provided to the research team. To date, five parents/legal guardians have opted out.

Ontario, it is not possible to access special education data through the Ministry of Education, making province-wide surveillance unfeasible. The catchment area is therefore restricted to southeastern Ontario[11] where the coordinating centre for NEDSAC is located. The seven public and Catholic school boards in that region provide minimal data[11] to NEDSAC on children aged less than 15 years diagnosed with ASD, including home-schooled children. The agency that delivers the provincially funded intensive behavioural intervention program in southeastern Ontario also participates in NEDSAC. However, only children on the moderate to severe end of the autism spectrum are eligible for this program. To ensure that most preschoolers with an ASD diagnosis are captured, the main referral and assessment centre in southeastern Ontario for children who have a suspected developmental condition also identifies cases to NEDSAC.

In these three regions, research ethics boards and agency policies require us to obtain consent from parents or legal guardians to contact them directly for more detailed information on their child's condition. The participating agencies mail information packages about the study to families. Parents or legal guardians who want to provide more detailed demographic and diagnostic information to NEDSAC return a consent form to the research team. A researcher then collects the data by telephone. If no consent form is returned, the minimal information provided by the agencies (i.e. date of birth, sex, prevalence year, diagnostic subgroup) allows us to include all cases in our prevalence estimates.

In Manitoba, Children's Special Services, a government program that supports children with special needs throughout the province (excluding those on native reserves, which fall under federal jurisdiction), identifies cases of ASD among children aged less than 18 years to NEDSAC. Data collection in Manitoba differs from the three other regions: agency staff at Children's Special Services provide the demographic and diagnostic information that is collected from parents or legal guardians who return a consent form in southeastern Ontario, Prince Edward Island, and Newfoundland and Labrador.

All children identified through the agencies mentioned above would have been diagnosed by a qualified health professional (e.g. developmental pediatrician, psychologist, psychiatrist). We confirmed the diagnosis in a sample of children within the four regions, using examiners who had obtained research reliability in administering the Autism Diagnostic Interview-Revised[17] (ADI-R) and the Autism Diagnostic Observation Schedule-Generic[18] (ADOS-G), two reference-standard tools for diagnosing ASD. Of 145 children assessed, 96.6% met the "autism" cut-off on the ADI-R or the "ASD" cut-off on the ADOS-G. The Metropolitan Atlanta Developmental Disabilities Surveillance Program reported a similarly high proportion of children with a previous ASD diagnosis who met surveillance criteria for having ASD (98%).[3]

In 2002, three tertiary provincial referral and assessment agencies in Vancouver[19] undertook a chart review to identify children with ASD aged less than 15 years. Information on diagnosed cases continued to be collected this way until 2007, at which time data collection for British Columbia ceased. From 2003 to 2006, the Developmental Clinic at Alberta Children's Hospital in Calgary and various community sources in that city also provided data to NEDSAC on children with ASD aged less than 15 years. Although the British Columbia and Calgary data are not used to estimate prevalence because of incomplete case capture in those regions, they contribute to our understanding of the epidemiology of ASD in Canada (see *Correlates of age at diagnosis of autism spectrum disorders in six Canadian regions* in this issue).[20]

In both Prince Edward Island and Newfoundland and Labrador, the potential for identifying an individual more than once (which could result in duplicate records in the database and hence an overestimation of the number of children with ASD) is minimal. In Prince Edward Island, only one agency provides data to NEDSAC. In Newfoundland and Labrador, representatives from the two participating agencies meet to resolve potential duplicates prior to sending the information to the NEDSAC coordinating centre.[111] In Manitoba, although only one agency participates in NEDSAC, seven regional offices complete data collection forms. Thus, if a family moves within the province, two regional offices could potentially submit data collection forms for the same child. The chance that a child is identified more than once is highest in southeastern Ontario, where multiple agencies participate in NEDSAC.

Regardless of the region, when information on a new case is entered in the database, an algorithm searches for records from the same region with matching date of birth (including reversed day and month of birth) and first two letters of the child's surname and first name[111] (including transposed letters; i.e. if "abcd" is entered, the algorithm searches for exact matches and "cdab"). If potential duplicates are detected at this stage, the reporting agencies are contacted to ascertain whether the information they provided is accurate. If the reporting agencies confirm no errors in the information provided and it is reasonable to assume that the information refers to the same child,[111] no new record is created and the existing record is tagged as a suspected duplicate case.

[11] Includes the six counties of Hastings, Prince Edward, Lennox & Addington, Frontenac, Leeds & Grenville and Lanark.

[11] Date of birth, sex, prevalence year (year in which the child was first known to have an ASD diagnosis and reside in the surveillance region), diagnostic subgroup.

[20] The Provincial Autism Resource Centre, Department of Pediatrics, Sunnybill Health Centre for Children; The Provincial Programme in Medical Genetics, Department of Medical Genetics; and The Division of Child and Adolescent Psychiatry, Department of Psychiatry, BC Children's and Women's Health Centre, University of British Columbia.

[111] Individuals' names are not shared between departments during this process.

[111] "xxxx" is entered for cases from southeastern Ontario, Prince Edward Island, and Newfoundland and Labrador whose parents did not return a consent form.

[111] For example, the search algorithm finds an existing record with the same date of birth and first two letters of the surname and first name, the data are provided by two school boards in southeastern Ontario, and one of these reports that the child is no longer with that board.

Thus, when there is the possibility of a potential duplicate, only one record is created in NEDSAC.

NEDSAC now and in the future

Although data collection has ceased in British Columbia and Calgary, we continue to collect information on children with ASD in Manitoba, southeastern Ontario, Prince Edward Island, and Newfoundland and Labrador. We are currently analyzing the 2003–2008 data for these four regions. While inter-regional variations need to be interpreted with caution, given the different case ascertainment protocols, the findings will comprise the first population-based Canadian data on changes in ASD prevalence within a particular region over a six-year period. Agencies that provide services to individuals with ASD can use these data for planning and resource allocation.

In 2009, our group received funding from the Public Health Agency of Canada to evaluate the feasibility of linking datasets housed at the Manitoba Centre for Health Policy to supplement or replace our current surveillance protocol in that province. We are also collaborating with researchers in Quebec to explore the potential for linking datasets in that province to identify children with ASD. Various other initiatives, such as Population Data BC and the Child and Youth Data Lab in Alberta, could make population-based surveillance of ASD using administrative data a viable option in other areas of the country. The use of administrative data for surveillance purposes does present challenges in terms of data quality, as noted earlier. However, there are ways to deal with such challenges. For example, case definitions based on administrative data have been validated for conditions such as inflammatory bowel disease[20] and diabetes.[21] A group of investigators in Nova Scotia recently compared how accurately various combinations of diagnostic codes in three administrative health datasets identified children with ASD.[14] Using diagnoses made by the Autism Team at the IWK Health Centre in Halifax as the reference standard, the sensitivity ranged from 11.9% to 69.3% and the specificity from 77.3% to 97.7%. Linking education datasets with health ones, rather than relying on health sources alone, would likely improve detection rates (i.e. sensitivity).[9,10,14]

Conclusion

Linking administrative datasets is a cost-effective option that could allow us to expand ASD surveillance to more regions of the country. In light of the public health importance of this group of disorders,[5] NEDSAC will continue to evolve and to provide information for policy makers, families and advocates on the occurrence of ASD in Canada.

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Correlates of age at diagnosis of autism spectrum disorders in six Canadian regions

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Abstract

Introduction: Early identification of autism spectrum disorders (ASD) is important, since earlier exposure to behavioural intervention programs may result in better outcomes for the child. Moreover, it allows families timely access to other treatments and supports.

Methods: Using generalized linear modeling, we examined the association between child and family characteristics and the age at which 2180 children were diagnosed with ASD between 1997 and 2005 in six Canadian regions.

Results: A diagnosis of pervasive developmental disorder-not otherwise specified (PDD-NOS) or Asperger syndrome, rural residence, diagnosis in more recent years, and foreign birthplace were associated with a later age at diagnosis. Children who are visible minorities or who have siblings with ASD were more likely to be diagnosed earlier. Collectively, these factors explained little of the variation in age at diagnosis, however.

Conclusion: While it is encouraging that ethnocultural identity, neighbourhood income, urban or rural residence, and sex of the child were not major contributors to disparities in the age when children were identified with ASD, more work is needed to determine what does account for the differences observed. Regional variations in the impact of several factors suggest that aggregating data may not be an optimal strategy if the findings are meant to inform policy and clinical practice at the local level.

Keywords: *autism spectrum disorder, age at diagnosis, surveillance, Canada, Asperger syndrome, autistic disorder, pervasive child developmental disorder*

Introduction

The Centers for Disease Control and Prevention, the American Academy of Pediatrics and the Canadian Institutes of Health Research all highlight the importance of identifying children with autism spectrum disorders (ASD) at as young an age as possible.[1-3] A recent review describes the benefits of early diagnosis,[4] one of which is earlier access to intervention programs. These programs lead to improvements in intellectual functioning and adaptive behaviour.[5] Earlier exposure increases the likelihood that intervention "will alter the abnormal developmental trajectory of individuals with ASD and help guide brain and behavioural development back toward a normal pathway and, in some cases, prevent the full syndrome of ASD."[6] This "sooner is better" theory is supported by studies demonstrating that earlier intervention improves outcomes.[7,8] In Canada, the time it takes to diagnose ASD may also have implications in terms of funding for and hence access to treatment.

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For example, in British Columbia the government provides up to $22,000 each year to families who have a child with ASD until that child reaches six years of age, at which time funding is reduced to $6,000 per year.[9]

There are other benefits to early detection. Parents report feelings of relief when a diagnosis is made, as it gives them a better understanding of their child's behaviour.[10] A diagnosis also confers eligibility for services and supports and gives parents the option of seeking genetic counselling.

While an ASD diagnosis can often be made reliably when a child is aged between two and three years,[11,12] many children are not diagnosed until four years of age or more.[1,13,14] In light of this, national campaigns have been launched in both Canada and the United States (US) to promote earlier detection of ASD.[15,*]

The factors that contribute to variations in age at diagnosis of ASD have not been widely studied, although place seems to play a role. A recent study reported differences in the median age at diagnosis across four regions of Canada,[14] and a population-based study in the US reported differences across 13 surveillance sites in the age at which children were first identified with ASD.[16] Results for other factors are less consistent. In one study, boys were identified significantly earlier than girls,[16] whereas other studies have reported no sex differences in age at diagnosis.[17,22] Living in a rural area and lower household income were associated with a later age at diagnosis in one survey of 969 caregivers of children with ASD in Pennsylvania,[17] whereas no significant differences were detected for those same factors in an online survey completed by 146 caregivers of children with ASD in Virginia.[18] One study that reviewed Philadelphia Medicaid claims found that Caucasian children with autistic disorder were diagnosed significantly earlier than African-American children.[22] In contrast, the previously mentioned survey of 969 caregivers in Pennsylvania revealed no significant differences in the age at which Caucasian and ethnic minority children were diagnosed.[17] Similarly, data from the Metropolitan Atlanta Developmental Disabilities Surveillance Program reported no significant effects of race or ethnicity on timing of the ASD diagnosis.[13]

Since the majority of these studies were conducted in the US, it is important to replicate them in other areas with sufficiently large sample sizes to consider multiple factors simultaneously. The analysis presented in this paper used data collected through a Canadian ASD surveillance program. Our objective was to examine the association between child and family characteristics and the age at which children in six regions of Canada were first diagnosed with ASD between 1997 and 2005. A further objective was to explore whether there were any differential effects of the characteristics examined across regions and diagnostic subgroups.

Methods

Sample

The sample consisted of 2180 children from six regions of Canada whose information is recorded in the National Epidemiologic Database for the Study of Autism in Canada (NEDSAC). NEDSAC was established in 2001, and surveillance of ASD cases has been ongoing since 2002 among children aged less than 18 years in Manitoba and Prince Edward Island and since 2003 among children aged less than 15 years in southeastern Ontario[†] and Newfoundland and Labrador. In 2002, three provincial referral and assessment centres in Vancouver, British Columbia, undertook a chart review to identify ASD cases among children aged less than 15 years. Information on diagnosed cases continued to be collected through chart review until 2007, at which time data collection in British Columbia ceased. From 2003 to 2006, the Developmental Clinic at Alberta Children's Hospital in Calgary and various community sources in that city also provided data to NEDSAC on children with ASD aged less than 15 years. More detailed information about the regional case ascertainment and data collection protocols is available in a status report published in this issue.[23]

To compensate for the different start dates and target age groups in the surveillance regions, the sample included children born in 1989 or later who were first diagnosed with ASD before their fifteenth birthday. We also restricted the sample to those who were initially diagnosed between 1997 and 2005, inclusive; relatively few children were diagnosed prior to 1997, and the last complete year of data collection for Calgary was 2005. Figure 1 illustrates the sample selection process.

Analysis

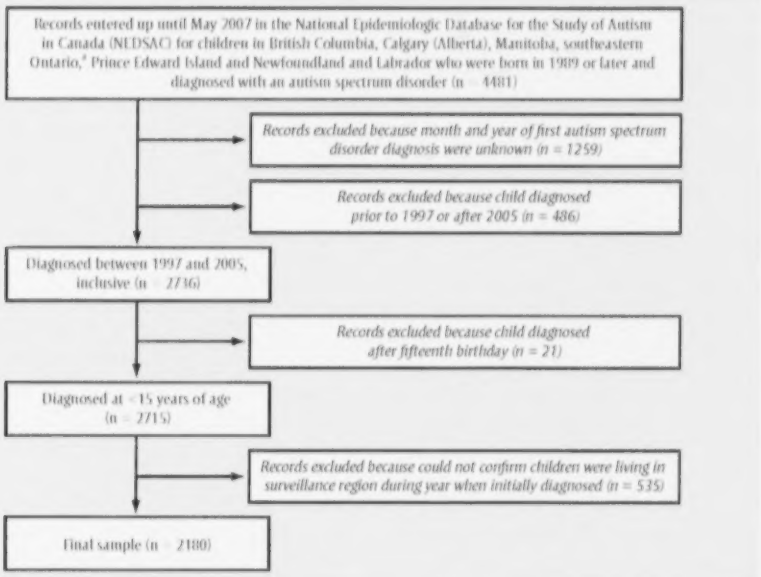
NEDSAC contains basic demographic and diagnostic information on children with ASD. We examined the following characteristics in terms of potential associations with age at diagnosis:

(1) diagnostic subgroup (autistic disorder/pervasive developmental disorder-not otherwise specified [PDD-NOS]/Asperger syndrome/autism spectrum disorder; the latter category was used when a general diagnosis of ASD was provided to the parent or recorded on the child's file);
(2) ethnocultural identity, either self-identified by parents or recorded on the child's file (Caucasian/visible minority/Aboriginal; when a child is a member of a non-Aboriginal visible minority and also identified as Aboriginal, he or she was classified as Aboriginal);
(3) being adopted (no/yes);
(4) neighbourhood median household income as a proxy measure of household income (lowest/middle/highest tertile, based on information from the Canadian Census for all private households in the area delineated by the first three characters of the postal code of last known residence in the surveillance region[24]);
(5) last known residence in the surveillance region (urban/rural, as defined by the second character of the postal code, where a "0" was coded as rural and all other numbers were coded as urban[25]);

[*] http://www.autismspeaks.org/press/cdc_awareness_campaign.php

[†] Includes the six counties of Hastings, Prince Edward, Lennox & Addington, Frontenac, Leeds & Grenville and Lanark.

FIGURE 1
Sample selection



[a] Includes the six counties of Hastings, Prince Edward, Lennox & Addington, Frontenac, Leeds & Grenville and Lanark.

(6) sex (boy/girl);
(7) three-year category for year of initial diagnosis (1997–1999/2000–2002/2003–2005);
(8) birthplace (Canada/other country); and
(9) whether any siblings have been diagnosed with ASD (no/yes).

Region was entered as a covariate in the models to control for its effects on age at diagnosis, but it was not one of the variables under investigation. Accordingly, we have not reported results for region in the tables or text. A priori, we decided to include only main effects in the regression models and did not test for interactions between the variables.

Generalized linear regression models were fit for the total sample using statistical package SAS version 9.1.3 (SAS Institute Inc.) and specifying the log link function and gamma distribution.[26] To reduce assumptions regarding the distribution of the dependent variable (age at diagnosis), we used the quasi-likelihood (or generalized estimating equation) approach instead of the full likelihood approach. The first model included only those observations where complete information was available ("complete-case model"). Some of the variables we examined had a large number of missing values (see Table 1; missing values ranged from none for diagnostic subgroup, sex and year of initial diagnosis to 22.9% for ethnocultural identity). Although imputation methods have their limitations, the current statistical literature recommends that missing values be imputed rather than restricting the analysis to cases with complete data.[27] We used the Markov chain Monte Carlo method[28] to generate 10 imputations for each missing data point. Imputed values were not rounded.[29] Generalized linear regression models were then fit to the imputed data and the final parameter estimates were summarized using the SAS PROC MIANALYZE procedure ("imputed models"). R^2 values, which in linear regression are used to indicate how much of the variation in the dependent variable is explained by the independent variables, were calculated using the following formula: 1 – (raw residual sum of squares)/(total sum of squares).[30]

We then fit separate regression models for the British Columbia and Manitoba data to examine whether the effects of the independent variables varied between those regions. The other four regions were not included in this analysis because the cell counts for certain variables were insufficient to produce meaningful parameter estimates. We also fit separate models for the three diagnostic subgroups ("autism spectrum disorder" was excluded from this analysis) using a forward stepwise variable selection procedure and specifying a p-value less than .10 for entering and retaining variables in the model.

All references to significance are based on two-tailed tests using an alpha of .05.

Results

Table 1 shows frequency distributions as well as mean and median ages at diagnosis for the independent variables. All variables were retained in the models for the total sample and for the British Columbia and Manitoba subsamples, as the variance inflation factors and condition indices were below values indicating potential problems with multicollinearity.[31,32] Tables 2 and 3 give the exponentiated parameter estimates from the regression models, with the significant findings bolded. Each estimate is the ratio of the expected age at diagnosis relative to the reference category. For example, the exponentiated parameter estimate for Asperger syndrome is 1.72 (Table 2, imputed models). This indicates that children with Asperger syndrome were, on average, diagnosed 1.72 times later than children with autistic disorder (the reference category) when all the other variables were held constant.

For the total sample, a diagnosis of PDD-NOS or Asperger syndrome, rural residence (imputed models), more recent diagnosis, and foreign birthplace were significantly associated with a later age at diagnosis. Conversely, children from visible minority groups or with a sibling with ASD were diagnosed significantly earlier (Table 2). Most of the variables that were significant for the total sample remained significant for the British Columbia subsample, apart from living in a rural area (Table 3). Only PDD-NOS, Asperger syndrome and Aboriginal identity (imputed models) were significantly associated with age at diagnosis for the Manitoba subsample (Table 3). When models were fit for the diagnostic

TABLE 1
Frequency distribution and mean and median ages at diagnosis for independent variables included in multiple regression analyses to examine associations with age at first diagnosis of an autism spectrum disorder (ASD)

Independent variables	Total sample (N = 2180)		Age at diagnosis	
	Number, n	Frequency, %	Mean, months (SD)	Median, months (IQ range)
Total	2180	100.0	60.8 (32.4)	50.0 (37.0)
Diagnostic subgroup				
Autistic disorder[a]	852	39.1	57.7 (31.9)	48.0 (29.0)
PDD-NOS[b]	320	14.7	69.2 (32.3)	60.0 (44.8)
Asperger syndrome	164	7.5	94.0 (29.8)	92.0 (46.5)
ASD[c]	844	38.7	54.3 (28.8)	45.0 (27.0)
Ethnocultural identity				
Caucasian	1142	52.4	60.3 (32.6)	50.0 (38.0)
Visible minority[d]	467	21.4	57.7 (30.3)	47.0 (28.0)
Aboriginal[e]	72	3.3	67.7 (36.2)	52.5 (55.5)
Unknown	499	22.9	63.9 (33.2)	53.0 (42.0)
Adopted				
No	2101	96.4	60.4 (32.0)	50.0 (36.0)
Yes	58	2.7	75.6 (42.1)	64.0 (77.5)
Unknown	21	1.0	58.7 (34.0)	48.0 (34.5)
Neighbourhood median household income[f]				
Lowest tertile	741	34.0	59.5 (31.7)	48.0 (36.0)
Middle tertile	738	33.9	61.4 (32.4)	51.0 (37.0)
Highest tertile	683	31.3	61.5 (33.1)	51.0 (39.0)
Unknown	18	0.8	63.9 (38.7)	55.0 (42.5)
Last known residence in surveillance region[g]				
Urban	1835	84.2	60.6 (32.5)	50.0 (38.0)
Rural	336	15.4	61.5 (31.8)	51.0 (37.8)
Unknown	9	0.4	71.4 (50.8)	52.0 (76.5)
Sex				
Boy	1809	83.0	60.6 (32.1)	50.0 (36.0)
Girl	371	17.0	61.8 (34.0)	49.0 (39.0)
Year of initial diagnosis, three-year category				
1997–1999	363	16.7	50.4 (19.8)	46.0 (23.0)
2000–2002	796	36.5	57.7 (30.3)	48.5 (34.0)
2003–2005	1021	46.8	66.9 (36.2)	54.0 (49.0)
Country of birth				
Canada	1867	85.6	58.2 (30.6)	48.0 (32.0)
Other	76	3.5	83.0 (40.7)	73.0 (72.8)
Unknown	237	10.9	74.0 (37.7)	63.0 (55.5)
Sibling(s) with an ASD				
No	1853	85.0	61.4 (32.5)	51.0 (38.0)
Yes	209	9.6	53.2 (28.5)	46.0 (29.5)
Unknown	118	5.4	65.5 (36.9)	50.0 (55.0)

Continued on the following page

TABLE 1 (Continued)
Frequency distribution and mean and median ages at diagnosis for independent variables included in multiple regression analyses to examine associations with age at first diagnosis of an autism spectrum disorder (ASD)

Independent variables	Total sample (N = 2180)		Age at diagnosis	
	Number, n	Frequency, %	Mean, months (SD)	Median, months (IQ range)
Region				
British Columbia	1247	57.2	64.7 (34.0)	54.0 (43.0)
Calgary, Alberta	180	8.3	54.1 (28.3)	46.0 (34.0)
Manitoba	493	22.6	56.2 (29.8)	47.0 (29.0)
Southeastern Ontario[h]	116	5.3	63.8 (32.8)	54.0 (51.0)
Prince Edward Island	54	2.5	57.7 (31.5)	45.5 (37.5)
Newfoundland and Labrador	90	4.1	43.3 (18.6)	39.0 (16.5)

Abbreviations: ASD, autism spectrum disorder; IQ, interquartile; NEDSAC, National Epidemiologic Database for the Study of Autism in Canada; PDD-NOS, Pervasive developmental disorder not otherwise specified; SD, standard deviation.

[a] Includes children diagnosed with childhood autism and infantile autism.

[b] Includes children diagnosed with childhood onset pervasive developmental disorder, atypical autism and other pervasive developmental disorders.

[c] General diagnosis of autism spectrum disorder was provided to parents or recorded on child's file.

[d] Excludes Aboriginal identity.

[e] First Nations/Native American, Inuit or Métis. In Manitoba, the on-reserve population is not included in NEDSAC. Reserves are under federal jurisdiction and are not served by the agency in Manitoba that identifies cases to NEDSAC.

[f] Based on the median household income for all private households in the area delineated by the first three characters of the postal code.[24]

[g] Based on the second character of the postal code, where 0 was coded as rural and all other numbers were coded as urban.[25]

[h] Includes the six counties of Hastings, Prince Edward, Lennox & Addington, Frontenac, Leeds & Grenville and Lanark.

subgroups, the only variable that was consistently significant was year of diagnosis. Children in the latter two-thirds of the study period were diagnosed significantly later than children diagnosed between 1997 and 1999 (data not shown). The other variables that showed significant associations in the diagnostic subgroup analysis included the following: being adopted for autistic disorder (point estimate = 1.21; 95% CI: 1.01–1.46) and PDD-NOS (point estimate = 1.42; 95% CI: 1.11–1.83); foreign birthplace for autistic disorder (point estimate = 1.45; 95% CI: 1.22–1.72); and having a sibling with ASD for PDD-NOS (point estimate = 0.79; 95% CI: 0.67–0.93).

Discussion

Diagnostic subgroup has been associated with age at diagnosis of ASD in several studies.[13,17,33,34] In our sample too, children with Asperger syndrome or PDD-NOS were generally diagnosed later than children with autistic disorder (Tables 2 and 3). Variations in the age at diagnosis among subgroups may be due to differences in the severity of core ASD symptoms: children with autistic disorder typically have more social, communication and cognitive delays than children with PDD-NOS or Asperger syndrome,[34] which may elicit earlier concerns on the part of parents or professionals.

We found significant associations between ethnocultural identity and age at diagnosis. Little is known about the influence of cultural factors on when children with ASD are diagnosed. A recent Dutch study reported that ethnic minorities are underrepresented in terms of referrals to ASD assessment centres.[35] The authors noted that pediatricians may attribute social and communication delays among these children to cultural factors.[35] Such an explanation is not consistent with our findings of earlier diagnosis among visible minorities in the total sample (Table 2) and in the subsample of cases from British Columbia (Table 3); however, it could partially explain why Aboriginal children tended to be diagnosed later than Caucasian children in Manitoba (Table 3). Mandell et al. found that the impact of ethnicity on age at diagnosis was more pronounced for children on the wider autism spectrum (i.e. those diagnosed with Asperger syndrome or PDD-NOS).[19] We did not include product terms in the multiple regression models, but we found no significant effects of ethnocultural identity on age at diagnosis when we stratified by diagnostic subgroup (data not shown). Thus, in our sample, the influence of ethnocultural identity on age at diagnosis did not appear to be concentrated at one end of the autism spectrum.

It has also been suggested that the symptoms of ASD in adopted children are apt to be mistakenly attributed to early childhood experiences, thus delaying the diagnosis.[17] Although we found no association between adoption and age at diagnosis in the total sample or in the British Columbia and Manitoba subsamples, it was significant for the autistic disorder and PDD-NOS subsamples. Thus, future studies that examine factors related to timing of diagnosis should consider including this variable.

In contrast to several American studies,[17,21] we found no association between income and age at diagnosis in our sample (apart

TABLE 2
Ratio of expected age at diagnosis of an autism spectrum disorder (ASD) compared to reference category, for total sample

Independent variables	Complete-case model (n = 1506; R^2 = .22)		Imputed models (N = 2180; R^2 = .21[a])	
	Exponentiated parameter estimate (95% CI)	*p*-value	Exponentiated parameter estimate (95% CI)	*p*-value
Diagnostic subgroup				
Autistic disorder	—	—	—	—
PDD-NOS	**1.22 (1.14–1.31)**	**<.0001**	**1.26 (1.19–1.34)**	**<.0001**
Asperger syndrome	**1.75 (1.63–1.89)**	**<.0001**	**1.72 (1.62–1.83)**	**<.0001**
ASD	0.99 (0.94–1.04)	.707	0.97 (0.93–1.02)	.248
Ethnocultural identity				
Caucasian	—	—	—	—
Visible minority	**0.90 (0.85–0.96)**	**.001**	**0.90 (0.85–0.95)**	**<.0001**
Aboriginal	1.06 (0.95–1.18)	.304	1.11 (0.99–1.24)	.070
Adopted				
No	—	—	—	—
Yes	0.93 (0.73–1.18)	.556	1.11 (0.98–1.27)	.109
Neighbourhood median household income				
Lowest tertile	—	—	—	—
Middle tertile	1.00 (0.94–1.05)	.944	1.02 (0.98–1.08)	.325
Highest tertile	1.00 (0.94–1.06)	.899	1.03 (0.98–1.08)	.314
Last known residence in surveillance region				
Urban	—	—	—	—
Rural	1.05 (0.98–1.12)	.170	**1.07 (1.01–1.14)**	**.025**
Sex				
Boy	—	—	—	—
Girl	1.05 (0.98–1.12)	.132	1.02 (0.96–1.07)	.591
Year of initial diagnosis, three-year category				
1997–1999	—	—	—	—
2000–2002	1.05 (1.00–1.11)	.070	**1.09 (1.04–1.15)**	**.001**
2003–2005	**1.23 (1.16–1.30)**	**<.0001**	**1.25 (1.19–1.31)**	**<.0001**
Country of birth				
Canada	—	—	—	—
Other	**1.36 (1.20–1.55)**	**<.0001**	**1.39 (1.22–1.58)**	**<.0001**
Sibling(s) with an ASD				
No	—	—	—	—
Yes	**0.91 (0.85–0.98)**	**.008**	**0.90 (0.84–0.96)**	**.003**

Abbreviations: ASD, autism spectrum disorder; CI, confidence interval; PDD-NOS, Pervasive developmental disorder not otherwise specified.

Note: Reference categories are indicated by the long dash. Bolding indicates a statistically significant association based on $p < .05$.

[a] Average R^2 for 10 datasets with values imputed for missing data points.

from the imputed models for British Columbia, where the association was in the opposite direction from what might be expected: children living in neighbourhoods with the highest median incomes tended to be diagnosed later than children from neighbourhoods with the lowest median incomes). There are two possible explanations for the difference between our findings and those of the American studies. First, the latter used individual-level measures of income, whereas we used a neighbourhood-level measure as a proxy for household income. Second, it is possible that we found no association because there are generally fewer financial barriers to accessing health services in Canada than in the US.

Mandell et al. also reported that children from rural areas tended to be diagnosed with ASD later than children living in urban areas.[17] They hypothesized that a higher population density contributes to a critical mass of children with ASD, leading to greater familiarity with the disorder on the part of health professionals and families and hence earlier recognition.

TABLE 3
Ratio of expected age at diagnosis of an autism spectrum disorder (ASD) compared to reference category for British Columbia and Manitoba subsamples

	British Columbia				Manitoba			
	Complete-case model (n = 847; R^2 = .15)		Imputed models (n = 1247; R^2 = .15[a])		Complete-case model (n=306; R^2= .30)		Imputed models (n = 493; R^2 = .35[a])	
	Exponentiated parameter estimate (95% CI)	*p*-value	Exponentiated parameter estimate (95% CI)	*p*-value	Exponentiated parameter estimate (95% CI)	*p*-value	Exponentiated parameter estimate (95% CI)	*p*-value
Diagnostic subgroup								
Autistic disorder	—	—	—	—	—	—	—	—
PDD-NOS	**1.14 (1.03–1.26)**	**.012**	**1.16 (1.07–1.26)**	**<.0001**	**1.52 (1.33–1.74)**	**<.0001**	**1.62 (1.44–1.83)**	**<.0001**
Asperger syndrome	**1.62 (1.45–1.81)**	**<.0001**	**1.51 (1.37–1.66)**	**<.0001**	**1.86 (1.62–2.14)**	**<.0001**	**2.00 (1.79–2.23)**	**<.0001**
ASD	0.95 (0.88–1.02)	.179	0.95 (0.89–1.01)	.087	1.05 (0.94–1.16)	.408	1.03 (0.95–1.13)	.483
Ethnocultural identity								
Caucasian	—	—	—	—	—	—	—	—
Visible minority	**0.87 (0.82–0.94)**	**.0001**	**0.86 (0.81–0.92)**	**<.0001**	0.96 (0.86–1.09)	.554	0.93 (0.83–1.05)	.258
Aboriginal	1.00 (0.83–1.20)	.986	1.07 (0.89–1.29)	.471	1.17 (1.00–1.37)	.056	**1.16 (1.01–1.33)**	**.035**
Adopted								
No	—	—	—	—	—	—	—	—
Yes	0.83 (0.56–1.22)	.331	1.12 (0.94–1.34)	.198	1.10 (0.75–1.60)	.634	1.13 (0.92–1.37)	.239
Neighbourhood median household income								
Lowest tertile	—	—	—	—	—	—	—	—
Middle tertile	1.04 (0.96–1.12)	.374	1.07 (1.00–1.14)	.061	0.92 (0.82–1.03)	.130	0.97 (0.88–1.06)	.495
Highest tertile	1.04 (0.96–1.13)	.326	**1.08 (1.01–1.15)**	**.029**	0.94 (0.83–1.07)	.369	1.03 (0.93–1.14)	.624
Last known residence in surveillance region								
Urban	—	—	—	—	—	—	—	—
Rural	1.02 (0.89–1.18)	.731	1.07 (0.96–1.19)	.208	1.08 (0.97–1.21)	.177	1.09 (0.99–1.20)	.093
Sex								
Boy	—	—	—	—	—	—	—	—
Girl	1.00 (0.92–1.10)	.939	0.97 (0.90–1.04)	.390	1.08 (0.94–1.23)	.273	1.05 (0.94–1.17)	.374
Year of initial diagnosis, three-year category								
1997–1999	—	—	—	—	—	—	—	—
2000–2002	1.05 (0.97–1.14)	.203	**1.12 (1.05–1.21)**	**.001**	0.98 (0.87–1.10)	.740	1.03 (0.92–1.15)	.654
2003–2005	**1.28 (1.18–1.38)**	**<.0001**	**1.33 (1.24–1.42)**	**<.0001**	1.08 (0.96–1.22)	.202	1.08 (0.98–1.20)	.117
Country of birth								
Canada	—	—	—	—	—	—	—	—
Other	**1.38 (1.20–1.58)**	**<.0001**	**1.38 (1.22–1.57)**	**<.0001**	1.28 (0.87–1.90)	.210	1.28 (0.85–1.92)	.229
Sibling(s) with an ASD								
No	—	—	—	—	—	—	—	—
Yes	0.89 (0.80–1.00)	.052	**0.83 (0.75–0.92)**	**<.0001**	0.96 (0.85–1.08)	.473	1.01 (0.88–1.15)	.914

Abbreviations: ASD, autism spectrum disorder; CI, confidence interval; PDD-NOS, Pervasive developmental disorder not otherwise specified.

Note: Reference categories are indicated by the long dash. Bolding indicates a statistically significant association based on $p < .05$.

[a] Average R^2 for 10 datasets with values imputed for missing data points.

Children from rural areas may also face greater barriers in accessing specialized diagnostic services. If the latter is a strong determinant of diagnostic delays, one might expect to see an association between rural status and age at diagnosis in Manitoba: it has a land area of almost 650 000 km^2 and the two main referral and assessment centres for children with suspected ASD are in Winnipeg. However, while children in rural areas tended to be diagnosed later than those in urban areas in all the models, the association was only significant in the imputed models for the total sample. Future studies could examine the distance to specialized referral and assessment centres instead of urban versus rural residence, something we could not do because our database does not contain addresses.

With a larger number of girls than boys in our sample (n = 371) and the lack of an association between sex and age at diagnosis across all analyses, we found no evidence of what Shattuck et al. referred to as "sex bias in cultural expectations of children's behaviour or in clinical practices for screening, referral and diagnosis."[16] A less encouraging finding is that the age at diagnosis increased over the study period in the total sample, although the data from British Columbia, the region with the largest sample size, may have been the prime driver of this trend; no significant increase was detected in Manitoba (Table 3). (A temporal increase in the age at diagnosis of ASD has also been reported for southeastern Ontario.[14]) Children were diagnosed about 25% later in 2003 to 2005 compared to 1997 to 1999 (Table 2). A similar pattern was seen across all diagnostic subgroups (data not shown), and therefore it is unlikely that this finding can be attributed solely to increased referrals of older children with milder symptoms in the later years of the study period. It may be that assessment services in some regions were becoming overburdened due to increased referrals over the study period, resulting in longer wait times. While we do not have the data to examine this hypothesis, the Standing Senate Committee on Social Affairs, Science and Technology, in its 2007 report on the enquiry into funding ASD treatment in Canada, noted that parents of children with ASD frequently have difficulty accessing assessment and diagnostic services in a timely manner.[16]

Two factors we examined—birthplace and having a sibling with ASD—have not, to our knowledge, been included in other studies of age at diagnosis of ASD. Children born in another country were more likely to be diagnosed at a later age than Canadian-born children. This variable had one of the largest effects, second only to a diagnosis of Asperger syndrome (Table 2). It would be interesting to study this association in a larger sample with more discrete categories for birthplace. The findings could provide a basis for recommendations to improve screening and assessment services among groups that may be at risk for later identification, thus ensuring that all children with ASD are diagnosed as early as possible.

Having a sibling with ASD was associated with an earlier age at diagnosis in the total sample and in British Columbia (Tables 2 and 3), although this variable was only significant for the PDD-NOS subgroup. This suggests that the milder symptoms of PDD-NOS may elicit earlier concerns if the child has a sibling who has already been diagnosed with ASD, whereas the symptoms of autistic disorder may be severe enough that parents or professionals are likely to become concerned early on regardless of whether there is another child with ASD in the family. If this hypothesis is correct, one would expect this variable to be significant for Asperger syndrome as well; however, it was not. This may be due to the small number of children in that group who had a sibling with ASD (n = 16). To better evaluate this hypothesis, it would be useful in future studies to include information on whether siblings are older or younger (we did not collect this information in NEDSAC prior to 2009).

One of our objectives was to determine whether the factors we examined for the total sample had a differential effect on age at diagnosis at the regional level. The only significant associations common to both British Columbia and Manitoba were for diagnostic subgroup (Table 3). This underscores the need to consider how aggregating data may obscure regional differences. If the findings from similar analyses are meant to inform policy and clinical practice at the local level, analytic strategies should be employed with this goal in mind. This is particularly relevant in Canada, where diagnostic services vary widely across jurisdictions. For example, in 2003 the provincial government in British Columbia established a province-wide network of clinicians who use standardized guidelines to assess and diagnose children and youth suspected of having ASD. In contrast, the main referral and assessment centres in Manitoba are located in Winnipeg. Thus, it is not surprising that the factors that influence when a child is diagnosed with ASD differ between these provinces.

The R^2 values indicate that the variables we examined accounted for little of the variation in age at diagnosis (15%–35%; Tables 2 and 3). In some ways, this is reassuring; it suggests that sociodemographic and socio-economic characteristics do not greatly influence when a child is diagnosed with ASD in Canada. It does mean, however, that more work needs to be done to identify what does account for the observed differences in age at diagnosis. Mandell et al. regressed a large number of individual-level variables on age at diagnosis, including ASD symptoms; whether the child had an intellectual disability, hearing impairment or seizures; how many physicians were seen before the diagnosis; whether developmental tests were conducted; and whether there was a referral to a specialist.[17] In their model, 46% of the variation in age at diagnosis remained unaccounted for. At least two studies have examined the influence of area-level factors on age at diagnosis.[19,21] In both these studies, most of the variation in age at diagnosis was associated with differences at the individual level. However, the area-level factors examined were primarily socio-economic and sociodemographic in nature. In future studies, we would like to explore whether health system characteristics are major determinants of when children with ASD are diagnosed. Such characteristics might include the number of developmental pediatricians per capita, the average age of pediatricians practicing in the area, whether practice parameters are in place for screening for ASD, and average wait times for assessment.

Limitations and strengths

One limitation of this study is the lack of clinical data on cognitive status, comorbidities and ASD symptoms, which have been shown in several US studies to be significantly associated with age at diagnosis.[16,17] Furthermore, while our surveillance protocols were designed to capture most children diagnosed with ASD in Manitoba, southeastern Ontario, Prince Edward Island, and Newfoundland and Labrador, the data from British Columbia and Calgary are primarily clinic-based, and therefore the extent of our case ascertainment in those regions is unknown. Another potential limitation concerns the multiple imputation procedure in SAS, which assumes that values are missing at random. It is not usually possible to test

this, but erroneous assumptions in this regard may have a minor impact on parameter estimates and standard errors.[37] The Markov chain Monte Carlo algorithm assumes multivariate normality to impute missing values. It has been demonstrated, however, that it generally performs well for categorical variables if the imputed values are not rounded,[39] which was the approach we used.

As shown in Figure 1, we excluded cases where the month and year of initial diagnosis were unknown (thus preventing us from calculating age at diagnosis) and cases where residence in the surveillance regions during the year of initial diagnosis could not be confirmed. We do not know what proportion of these cases would have met the criteria for inclusion in the sample; however, there is no reason to suspect that children who were diagnosed from 1997 to 2005 and who were not included in this analysis for any of the preceding reasons differed systematically from the sample in terms of both the independent variables and the age at which they were diagnosed with ASD. Nevertheless, we cannot rule out the possibility of bias in our estimates. Accordingly, this work needs to be replicated in other samples; our findings can help guide such research.

The strengths of this study include the large sample size, which provided good statistical power to examine the association between a number of factors and the age at which children were diagnosed with ASD. To our knowledge, this is the first study to examine foreign birthplace (for the child) and the presence of a sibling with ASD in relation to age at diagnosis, and it is the first analysis of its kind in a Canadian population. The large sample sizes for British Columbia and Manitoba allowed us to conduct separate analyses for those two regions, which revealed some potentially important differences in how certain factors relate to age at diagnosis at the regional level. Another strength of our analysis is that we only included children who resided in the surveillance regions at the time of diagnosis. This increases the probability that the findings reflect the local diagnostic situation during the study period.

Perhaps one of the most intriguing findings is how little of the variation in age at diagnosis was accounted for by our models. While this suggests that the socioeconomic and sociodemographic factors we examined had little impact on when children with ASD were identified—whereas a recent American study found that such factors were generally stronger predictors of age at diagnosis than symptom severity[13]—it does underscore the need to gain a better understanding of what contributes to disparities in age at diagnosis. Future studies should include more detailed information on the variables we examined, as well as a broader range of factors. These might include individual-level characteristics, such as the presence of clinical comorbidities that could conceivably delay recognition of the behavioural symptoms of ASD,[38] as well as health system characteristics, such as waiting times for referral and assessment. Such studies are crucial for ensuring earlier access to treatment and supports for all children with ASD and their families.

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The development of national indicators for the surveillance of osteoporosis in Canada

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Abstract

Introduction: The Public Health Agency of Canada, in collaboration with bone health and osteoporosis experts from across Canada (n = 12), selected a core set of indicators for the public health surveillance of osteoporosis using a formal consensus process.

Methods: A literature review identified candidate indicators that were subsequently categorized into an osteoporosis-specific indicator framework. A survey was then administered to obtain expert opinion on the indicators' public health importance. Indicators that scored less than 3 on a Likert scale of 1 (low) to 5 (high) were excluded from further consideration. Subsequently, a majority vote on the remaining indicators' level of public health importance was sought during a face-to-face meeting.

Results: The literature yielded 111 indicators, and 88 were selected for further consideration via the survey. At the face-to-face meeting, more than half the experts considered 39 indicators to be important from the public health perspective.

Conclusion: This core set of indicators will serve to inform the development of new data sources and the integration, analysis and interpretation of existing data into surveillance products for the purpose of public health action.

Keywords: osteoporosis, bone diseases, health status indicators, population surveillance, public health, consensus

Introduction

Public health surveillance is a core component of the Public Health Agency of Canada's (PHAC) mandate.[1] Regular surveillance of chronic conditions, conducted by PHAC's Centre for Chronic Disease Prevention and Control (CCDPC), is essential for providing the evidence to develop, implement, enhance and evaluate chronic disease prevention and management strategies. The CCDPC collaborates with regional, provincial/territorial, national and international governments and stakeholders to share knowledge of chronic disease in order to support policies, programs and public health interventions that aim to protect and improve the health of the Canadian population.[2]

Measures that reflect the health of a population or the performance of health care processes and outcomes are known as public health surveillance indicators.[3] There are a number of national initiatives that report on a limited number of indicators of chronic diseases;[4-7] however, a comprehensive indicator framework designed specifically for the public health surveillance of chronic diseases in Canada has not yet been described.

In consultation with PHAC's chronic disease surveillance advisory committees, the CCDPC developed a chronic disease surveillance indicator framework in 2007.[8] The framework categorizes indicators from established surveillance programs including those for arthritis, cancer, cardiovascular disease, chronic respiratory diseases, diabetes and mental illnesses into one of five dimensions: (1) individual risk and protective factors; (2) health status indicators; (3) health promotion and disease prevention indicators; (4) disease management indicators; and (5) environment-specific indicators. The main objectives of this indicator framework are to support PHAC's work on the surveillance of chronic diseases and to enhance federal, provincial/territorial and local/regional capacity to use, analyze and interpret surveillance data.

National surveillance of osteoporosis was initiated by CCDPC in 2008. Osteoporosis (i.e. thin or brittle bones) is a common skeletal disorder characterized by compromised bone strength that predisposes a person to fractures. According to the most recent estimates from PHAC, 1.5 million Canadians 40 years and older (10% of this population) reported being diagnosed with osteoporosis by a physician.[9] Osteoporosis is more prevalent among older individuals and also affects more women than men.[10] Its prevalence is projected to rise markedly over the next few decades as the number

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of older individuals increases.[12] The fractures associated with osteoporosis, specifically fractures of the spine and hip, are a significant cause of disability and mortality and a burden on health care utilization; however, there are evidence-based interventions that can substantially reduce the risk of these fractures.[13]

The primary objective of this study was to select a core set of indicators for the public health surveillance of osteoporosis in Canada. In the absence of indicators for a specific condition or disease, indicator developers rely on consensus-based processes.[14] We selected the proposed set of indicators through

(1) the development of an osteoporosis-specific indicator framework by tailoring the dimension descriptions that make up the CCDPC framework;
(2) a systematic rapid review of the literature to identify candidate indicators; and
(3) a formal consensus process, involving bone health and osteoporosis experts from across Canada, to select a core set of indicators.

The development of this core set of indicators for the surveillance of osteoporosis will inform the development of new data sources and support the integration, analysis and interpretation of existing data into surveillance products for dissemination. The regular monitoring and reporting of these indicators will help strengthen the evidence base that will ultimately inform future public health strategies and policies for preventing and managing osteoporosis in Canada.

Methods

Osteoporosis-specific indicator framework development

An osteoporosis-specific indicator framework was developed by tailoring the descriptions of the five indicator dimensions of the CCDPC Chronic Disease Indicator Framework in order to organize the candidate indicators that were extracted from the literature. Table 1 details the five dimensions of the framework specific to osteoporosis.

TABLE 1
Osteoporosis-specific indicator framework

Dimension	Description and examples
Individual protective/ risk factor indicators	Describe the individual factors (e.g. parental history of fragility/fracture), health behaviours (e.g. calcium and vitamin D intake), knowledge, attitudes, skills (e.g. knowledge of the benefits of weight-bearing exercise on bone health), and exposures (e.g. prolonged use of bone-depleting medication) that affect the risk of developing osteoporosis.
Health status indicators	Describe the magnitude (e.g. prevalence, incidence) and outcomes (e.g. quality of life, morbidity, mortality) of osteoporosis.
Health promotion/disease prevention indicators	Describe community- or population-based interventions (e.g. bone density screening programs, food fortification) that affect the development or management of osteoporosis.
Disease management indicators	Describe whether people are screened for and how people are managing their osteoporosis (e.g. use of bone-sparing medication, self-management).
Environment-specific indicators	Describe the broader physical (e.g. access to walking paths), social (e.g. food quality or availability) and economic factors (e.g. cost of living) that affect the development or management of osteoporosis.

Source: Adapted from Stewart, P. Chronic Disease Indicator Framework, Centre for Chronic Disease Prevention and Control, Public Health Agency of Canada; 2007 (unpublished).

Literature review

Data sources and searches. We conducted a systematic rapid review (i.e. a streamlined traditional systematic review)[15] of peer-reviewed and grey literature to identify candidate indicators for osteoporosis surveillance. For the peer-reviewed literature, we searched MEDLINE, Embase, Global Health and CINAHL. Our search strategy was developed in MEDLINE with the assistance of an experienced librarian scientist. The original search strategy was modified as required for the other databases. For the grey literature, we developed an Internet search strategy using the Google search engine. See Appendix 1 for the search strategies used.

Eligibility criteria and article selection. We searched peer-reviewed studies published between 1990 and 2009 that were population-based, descriptive or observational in design and described indicators for the surveillance of osteoporosis in an adult population. Our search of the grey literature took place on May 14th, 2009. Eligible records included population-based, descriptive or observational studies, reports or survey modules containing potential indicators for the surveillance of osteoporosis in an adult population. Only records published in English were considered.

Records retrieved from our search of the health sciences databases were screened for eligibility using the bibliographic record, that is, title, authors, keywords and abstract. We obtained the full text of those articles considered potentially relevant. Records retrieved from the Google search were screened for eligibility using the information provided on the related website and/or the full-text document (when available). The full-text records from both sources were retained for data extraction purposes when they met our eligibility criteria.

Data extraction. We developed a data extraction form based on the osteoporosis-specific indicator framework described in Table 1. Candidate indicators from the included records (peer-reviewed and non-peer-reviewed) were extracted and summarized using this form. We then reviewed and consolidated the list of indicators in order to eliminate any redundancy and classified the indicators into one of the five dimensions of the framework.

Consensus process

In order to select a core set of indicators from those retrieved from the literature, we used a two-step modified Delphi consensus-based process.[16] This included the use of an electronic survey and a face-to-face meeting with members of PHAC's Osteoporosis Surveillance Expert

Working Group. The Working Group was founded in 2008 by PHAC in collaboration with Osteoporosis Canada* in order to provide expert advice to the Agency on indicators, data sources and approaches to national surveillance of osteoporosis. It includes 10 clinician-researchers and 2 health scientists from across Canada with expertise in bone health and osteoporosis.

Step 1: Survey. Each member of the Working Group was emailed the survey and instructed to rank each candidate indicator according to its public health importance on a scale of 1 (low) to 5 (high).† In ranking an indicator for its public health importance, we asked the experts to consider (1) the size of the population that the indicator implicates or affects; (2) its importance in the prevention or management of osteoporosis; and/or (3) the severity of its potential outcome. After the experts had assigned each indicator a rank, they were asked to suggest population-based data sources (i.e. a registry or other data collection system that has information about all cases of a specific disease or injury in a geographically defined area that relates to a specific population) for a given indicator and comment on the quality of the data source. If an expert was unaware of a population-based data source, they were asked to comment on the feasibility of obtaining population-based data pertaining to the indicator.‡ Information regarding the availability and feasibility of the indicators was not used for evaluation purposes at this stage of the selection process. Lastly, the experts were asked to suggest any additional indicators for the surveillance of osteoporosis that they believed were important for monitoring the bone health and the impact of osteoporosis on Canadians. The experts were given ten days to complete the survey. Extensions were granted on an as-needed basis to maximize our response rate.

Following the synthesis of the survey results, candidate indicators with a median public health importance of 3 or more were retained for further consideration.§

Step 2: Face-to-face meeting. At the meeting, the experts were presented with the list of all the candidate indicators that had scored a median public health importance of 3 or more (as determined via the survey). Additional information presented for their consideration included the range in scores with respect to each indicator's public health importance (i.e. minimum and maximum) and relevant commentary about the potential data sources for a given indicator.

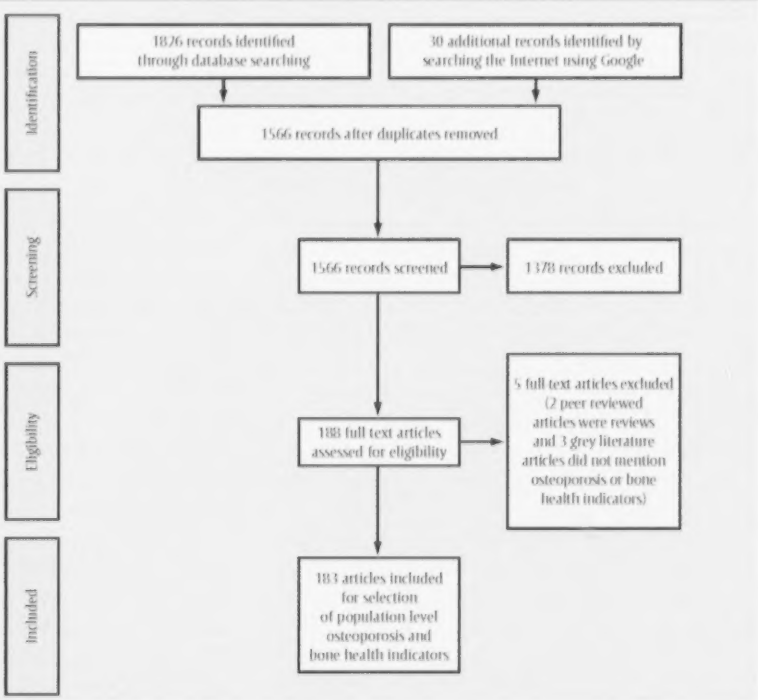
Following a review and open discussion of the survey results, the experts were asked to vote on the level of importance of the remaining indicators. The indicators that were rated by the majority (i.e. more than half) as having a high level of importance for inclusion were selected for the core set.

Results

Literature review

The literature review yielded 1826 peer-reviewed records and 30 records from the grey literature. A total of 183 records** met the eligibility criteria (see Figure 1 for flow diagram) and 111 unique candidate indicators were extracted from these and categorized according to the osteoporosis-specific indicator framework (see Table 1 and Figure 2).

FIGURE 1
Modified flow diagram showing systematic rapid review of literature to identify candidate indicators for osteoporosis surveillance



* http://www.osteoporosis.ca.

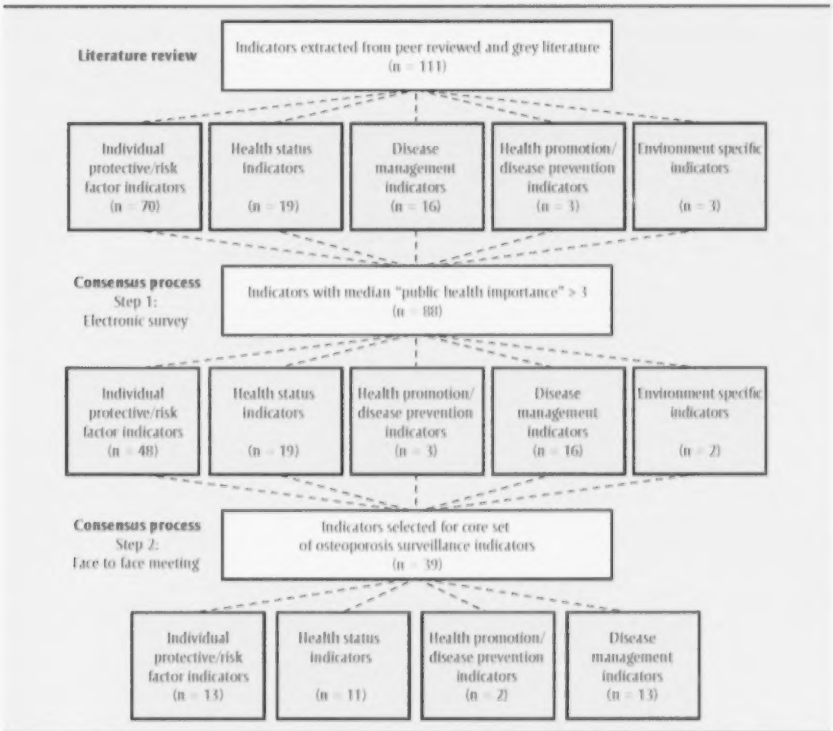
† Five-point Likert scale was adapted from a project by Majumdar et al. that developed a set of indicators for the evaluation of quality improvement efforts for adults with type 2 diabetes.[17]

‡ Ascertaining the feasibility of obtaining a given indicator was a criterion adapted from the Project for an Ontario Women's Health Evidence-Based Report (POWER) Study's Indicator Selection Criteria.[10]

§ Cut off point was adapted from a study by Majumdar et al. that developed a set of quality indicators for the evaluation of quality improvement efforts for adults with type 2 diabetes.[17]

** The list of included records from the systematic review is available on request.

FIGURE 2
Overview of study results



Consensus Process

Step 1: Survey. Eleven of the 12 members of the Osteoporosis Surveillance Expert Working Group completed the survey. Of the 111 candidate indicators extracted from the literature, 88 scored a median public health importance of 3 or more and were retained for further consideration (see Figure 1). The experts identified a dozen potential population-based data sources that could be used to measure these indicators (see Table 2).

Step 2: Face-to-face meeting. Nine out of the 12 members of the Working Group took part in the face-to-face consensus meeting. After an open discussion of the survey results, the majority considered 39 of the 88 candidate indicators to have a high level of importance: 13 individual protective and risk factors; 11 health status indicators; 2 health promotion and disease prevention indicators; and 13 disease management indicators (see Table 3). With respect to the remaining indicators, 28 had a medium and 19 had a low level of importance for inclusion in the core set.

Discussion

While there are several national initiatives on "health indicators,"[4,9] a comprehensive indicator framework for the surveillance of chronic diseases in Canada does not exist. This study represents the first step towards the development of a core set of indicators for the public health surveillance of osteoporosis in Canada. The 39 indicators selected through a formal consensus process cover all aspects of osteoporosis in the population including health promotion, risk and protective factors, health status, and disease management.

We felt it was important to document this process for the following reasons: (1) to address the lack of information on public health surveillance indicator development in the published literature; (2) to serve as a reference for developing surveillance indicators for other chronic conditions/diseases; and (3) to communicate the priority areas for PHAC's future data development efforts in osteoporosis surveillance.

In a like manner, the Australian Institute for Health and Welfare and the Data Working Group of the National Arthritis and Musculoskeletal Conditions Advisory Group developed a national set of consensus-based indicators (n = 16) that was guided by a conceptual framework for monitoring osteoarthritis, rheumatoid arthritis and osteoporosis.[19] Of this core set, 4 indicators were constructed to monitor the impact of osteoporosis: level of physical activity, osteoporosis prevalence, quality of life among those with osteoporosis and the number of hospitalizations for minimal trauma hip fractures.[19]

Future work will include developing operational definitions for each indicator including the rationale for its inclusion,

TABLE 2
Expert-identified population-based data sources for indicator development

Name of data source	Abbreviation
Canadian Multicentre Osteoporosis Study	CaMos
Canadian Community Health Survey	CCHS
Canadian Health Measures Survey	CHMS
Canadian Institute for Health Information	CIHI
Canadian Longitudinal Study on Aging	CLSA
Manitoba Bone Density Program	MBDP
Manitoba Centre for Health Policy	MCHP
Maximizing Osteoporosis Management in Manitoba	MOMM
Osteoporosis in Canada Report Card	OCRC
Ontario Drug Benefit Program	ODB
Recognizing Osteoporosis and Its Consequences in Quebec Programme	ROCQ
Régie de l'assurance maladie du Québec	RAMQ

TABLE 3
Core set of indicators for the surveillance of osteoporosis (n = 39)

Individual protective/ risk factor indicators (n = 13)	Health status indicators (n = 11)	Health promotion/disease prevention indicators (n = 2)	Disease management indicators (n = 13)
• Calcium intake	• Mortality attributable to hip fracture	• Community awareness regarding osteoporosis (e.g. falls prevention, vitamin D intake, physical activity)	• Rates of bone densitometry use
• Dairy intake	• Mortality attributable to osteoporotic fracture of any site	• Osteoporosis awareness media campaign	• Underwent osteoporosis testing after fragility fracture
• Vitamin D intake	• Mortality attributable to vertebral fracture/deformity		• Underwent osteoporosis treatment after fragility fracture
• General mobility	• Prevalence of bone mineral density outcomes		• Compliance with prescribed osteoporosis medications
• Height loss	• Prevalence of major osteoporotic fracture		• Taking prescription medications for osteoporosis (including bisphosphonates, calcitonin, sodium fluoride, selective estrogen receptor modulators or hormone replacement therapy)
• History of falls	• Prevalence of minor osteoporotic fracture		• Taking calcium or vitamin D supplements for osteoporosis
• Impaired balance	• Prevalence of vertebral deformity		• Number of prescriptions for osteoporosis
• Knowledge of protective factors for osteoporosis	• Prevalence/incidence/ diagnosis of osteoporosis		• Cost of acute hospital care for osteoporosis
• Knowledge of risk factors for osteoporosis	• Prevalence/Incidence of osteoporotic/fragility/low-energy fracture		• Cost of disability due to osteoporosis (value of activity days lost to short-term and long-term disability)
• Maternal and/or paternal and/or family history of hip fracture	• Quality of life (osteoporosis specific)		• Cost of physician care for osteoporosis
• Number of comorbid conditions	• Self-rated health		• Cost of drugs for osteoporosis
• Serum 25-hydroxycalciferol			• Cost of mortality due to osteoporosis (value of years of life lost due to premature death)
• Systemic steroid therapy			• Cost of post-acute care (e.g. rehabilitation)

the statistic or measure to be reported, the numerator and denominator to be used, existing or potential data sources and any notes, cautions or further instructions for calculating or interpreting results.[19] While the Osteoporosis Surveillance Expert Working Group identified several relevant national and provincial/territorial data sources (see Table 2), additional data sources will be warranted in order to populate all 39 indicators. Those indicators on which all provinces/territories could promptly collect information could form a minimum set that should be monitored and reported on regularly; for those indicators that cannot be reported on, data development should be undertaken to permit their eventual inclusion. Eventually, an evaluation of the indicators should be carried out to determine if the individual indicators meet quality criteria and if the set of indicators is comprehensive and meets decision and policy makers' information needs.[20]

Strengths and limitations

Despite the systematic approach we used to establish a core set of consensus-based indicators for osteoporosis surveillance, this study has several limitations. First, accelerating the timeframes of our literature review by conducting a systematic rapid review may have resulted in our missing some relevant information and biases.[15] To mitigate this possible limitation, the experts were able to suggest additional indicators that they believed important from a public health perspective in monitoring the bone health and the impact of osteoporosis on Canadians. Second, the modified Delphi consensus process relied on the opinions of a relatively small group (n = 12) of clinician-researchers and health scientists from across Canada. While there is no consensus regarding the method of selection, size and composition of an expert panel, the panel should reflect the full range of stakeholders who have an interest in the results of the study.[14] Lastly, while the indicators selected have face validity for measuring and tracking the impact of osteoporosis on Canadians, the indicators have yet to be operationalized and therefore their feasibility, accuracy and other characteristics are unknown.

Conclusion

A formal consensus-based process was used to incorporate evidence and expert opinion for the development of a core set of national indicators for the surveillance of osteoporosis. While current data gaps will influence the composition of this core set, the regular monitoring and reporting of the indicators that can be reported on, and the development of new data sources for those indicators that cannot be reported on, are important steps towards developing a stronger evidence base that

will ultimately inform future public health strategies and policies for preventing and managing osteoporosis in Canada.

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Appendix

APPENDIX 1 Search Strategies

MEDLINE (1990 to June 8, 2009)	
1	exp osteoporosis/ and exp data collection/ and exp public health/ and (nation* or population*).ti,ab.
2	limit 1 to (english language and humans and yr="1990-2009" and "all adult (19 plus years)")
Embase (1990 to 2009 week 23)	
1	exp osteoporosis/
2	exp "population and population related phenomena"/or exp disease surveillance/
3	(nation* or population*).ti,ab.
4	exp data collection method/ or exp mass screening/ or exp health survey/ or exp mathematical phenomena/
5	1 and 2 and 3 and 4
6	limit 5 to (yr=1990-2009 and human)
Global Health (1990 to May 2009)	
1	(nation* or population*).ti,ab.
2	osteopor*.ti.
3	(surveillanc* or survey* or screening* or questionnair* or data*).ti,ab.
4	1 and 2 and 3
5	limit 4 to yr=1990-2009
CINAHL (1990 to June 2009 week 3)	
1	TI(nation* or population*)
2	AB(nation* or population*)
3	TI(osteopor*)
4	TI(surveillanc* or survey* or screening* or questionnair* or data*)
5	AB(surveillanc* or survey* or screening* or questionnair* or data*)
6	(S1 or S2) and S3 and (S4 or S5)
	Google (1990 to May 14th, 2009)
	osteoporosis (national OR population) (indicators OR surveillance OR "health indices")

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Book review

Danse et santé : du corps intime au corps social

C. Couillard, PhD

Editors: Sylvie Fortin

Publisher: Presses de l'Université du Québec, Collection Santé et Société

Publication date: 2008

Number of pages: 330 pp.

Price: $30.00

ISBN: 978-2-7605-1543-7

This collection examines the intrinsic and complex relationship between health and dance. Meant primarily for balletomanes, including dancers, choreographers, coaches, researchers, physicians and journalists, among others, it includes rigorous academic analysis, presented in a detailed and informed way, as well as comments and descriptions of personal impressions, experiences and perceptions.

Danse et santé is divided into six parts, each made up of related articles in which the writers examine their perspective on health and dance. Giving a voice to these various contributors—all are involved with the world of dance—reinforces one of the central ideas of this work. Indeed, many of the texts suggest taking an approach whereby the dancers work with their colleagues to take charge of their own health and safety in the workplace. Others suggest that, although self-knowledge and kinesthesia go hand-in-hand with the aesthetic and artistic quest central to performance and creativity, listening to others and teamwork both play a major role in dancers' safety. Yet others point to the importance of government, unions and society in providing a framework for health, safety and injury prevention for performing artists.

The first part of *Danse et santé* describes the experiences of dancers and choreographers who strive constantly to balance artistic vision, aesthetic awareness and the limitations of their own or others' bodies. It also questions, in very clear terms, each and everyone's role, as well as that of the system, especially with regard to preventing injury. In the first chapter, Sylvie Fortin et al. address the issue of gender, referring to specific challenges in the areas of competitiveness, the creative process, training and even expectations.

The second part of the book describes the strategies used to inform the different actors of the opportunities to participate in the creative process, as well as the forms of power associated with each strategy. Pamela Newell and Sylvie Fortin detail the specific relationship between the choreographer and performer. The role of the performer in the creative process is part of a continuum between a traditional approach, in which the dancer's role is limited to reproducing movements, and a so-called decentralized role aimed at active and even spontaneous participation in the creation of the work. Linked to this continuum is the control the choreographer exercises over his or her work and that of the performer over his or her own body. The fourth and fifth chapters relate experiences in action research and in somatic education, and how students become increasingly aware of their bodies and question those automatically accepted assertions that determine issues of power over their own health.

The third part of *Danse et santé* provides an international perspective on injury prevention strategies in the dance world, beginning with a study on the diploma courses for dance teachers in France. Jill Green examines the concepts of health and well-being and related practices in America, including the medicalization of health and the benefits of (and issues connected with) alternative approaches such as somatic education that could become the dominant approach in the quest for an ideal. Blanka Rip et al. conclude this section by examining passion and the possible effects of obsessive passion on health and balance in a dancer. When passion is harmonious, it allows for flexibility and control; when it is obsessive, it affects everything, including health and injuries.

The fourth part of this book provides an interpretation of the professional dance milieu, examining the variety of structural

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factors that determine the health and safety of dancers in their work. Élise Ledoux et al. look at the organizational issue of business in dance, while Roger Hobden examines injury prevention during training.

The fifth part is the most unexpected. The eleventh chapter comprises works of fiction by four of the contributors who, based on personal accounts gathered through study and observation, re-create the dancer's world in poetry, a play, a diary and a narrative. The two other chapters in this section address the direct relationship between dance and illness. Christine Hanrahan and Nathalie Buisson, two dancers who have cancer, explain to Sylvie Fortin how dance helps them through their illness. Conversely, Aurore Després describes observing a dancer perform for hospital patients, thus giving them a gift of her art.

The sixth and final part gives various examples of the representation of illness and physical suffering through dance, often, but not exclusively, as a cathartic or auto-therapeutic process. The reflection on the physical form that begins in this contribution by Tamar Tembeck continues in the following chapter with a choreographic analysis of the definition and perception of the perfect body and of the expectations for achieving that ideal.

Overall, the contributors convincingly persuade us of the need for greater awareness of and control over the issues related to dancers' health and safety. An underlying theme is the matter of gender and the issues specific to female and male dancers, highlighted in particular by the rate of participation in the various workshops and studies (where there is a preponderance of women), as well as individual first-hand accounts. With a view to changing the sociocultural practices that have long prevented dancers from controlling their own bodies and their own health, the contributors advocate change among the dancers themselves, in their training and in society, to include stricter standards, healthier practices, revised expectations and new ways of thinking.

News

A new knowledge synthesis method that is applicable to public policies

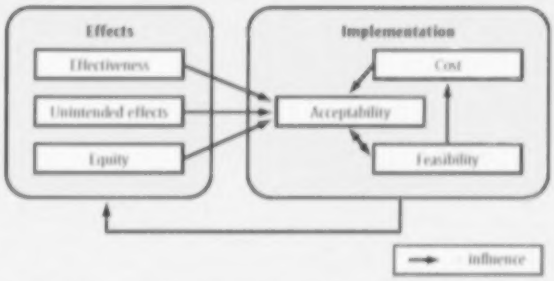
F. Morestin

The National Collaborating Centre for Healthy Public Policy (NCCHPP) has developed a method for synthesizing knowledge about public policies. This may be of interest to public health actors tackling chronic disease who are expected to provide comprehensive and contextualized evidence in order to inform policy making.

Some of the array of causes of chronic diseases have to be addressed through public policies. In this context, public health actors are called upon to produce knowledge syntheses in order to inform policy makers. But studying public policies raises specific challenges. Drawing inspiration from literature on the concept of evidence in public health, criteria involved in policy making, policy evaluation, deliberative processes as a means of collecting contextual knowledge, and methods proposed by established knowledge synthesis groups (such as the Community Preventive Services Task Force affiliated with the Centers for Disease Control and Prevention in the United States, the Cochrane Public Health Group in Australia, the National Institute for Health and Clinical Excellence in the United Kingdom), the NCCHPP has developed a method that is applicable to public policies.

This method guides rigorous knowledge syntheses on six dimensions relevant to the study of public policies (Figure 1): their effectiveness at preventing disease, their unintended effects, their equity-related issues (distribution of effects on different population groups), and their costs, feasibility and acceptability—three kinds of implementation issues of concern to policy makers.

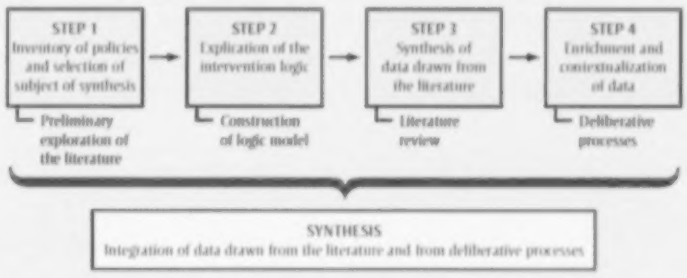
FIGURE 1
Relationships between the six dimensions for analyzing public policies



In order to gather knowledge on these different aspects in the most comprehensive and contextually relevant ways, the proposed process involves constructing the logic model of the policy under study, reviewing the scientific and grey literatures, and organizing deliberative processes that bring together relevant stakeholders to gather contextual information regarding the potential local implementation of this policy (Figure 2).

The NCCHPP has produced a guide that presents the method step-by-step and incorporates questions to ask oneself, practical advice, and several tools for

FIGURE 2
Steps in the knowledge synthesis process



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facilitating data collection and synthesis. The NCCHPP has also published a synthesis produced using this method, *Public Policies on Nutrition Labelling: Effects and Implementation Issues – A Knowledge Synthesis*; this document provides a concrete overview of how to use the proposed method and the kind of results that it can produce.

These documents are available in English and in French on the NCCHPP's website: http://www.ncchpp.ca/172/Publications.ccnpps. The NCCHPP also offers training material, workshops and methodological support to those who are interested in using this method.

With thanks to our 2011 peer reviewers

We are grateful to the following people for their significant contribution to *Chronic Diseases in Canada* and *Chronic Diseases and Injuries in Canada* as peer reviewers in 2011. Their expertise ensures the quality of our journal and promotes the sharing of new knowledge among peers in Canada and internationally.

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